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Dads and Dyads: stress and coping when a child has Retinoblastoma

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A thesis submitted in partial fulfillment of the requirements for the Doctor of Philosophy degree in Sociology

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Abstract

Child psychosocial oncology research offers limited examination of fathers' and dyadic stress and coping. Retinoblastoma (Rb) is a rare genetic eye cancer occurring at birth or early childhood. This qualitative sociological study examines individual and dyadic stress and coping across 4 fatherhood role categories when their child is diagnosed/treated for Retinoblastoma. Using purposive sampling, 23 Canadian Rb couples and 7 unmatched parents completed individual in-depth, semi-structured interviews. Findings confirm fatherhood role identity is diverse, influenced by the current situation, elements of discourse, and cultural references. Often contested in public and private spheres, fathering roles show transitional or permanent change tied to circumstance and dyadic support. In a stress process model, fathers primarily relied on problem-oriented and instrumental coping. Partners were the primary mediator of stress for all fathers, providing extensive emotional and informational supports. Using a systemic-transactional model of coping, most study dyads used positive coping strategies and were often supported by the extended clinical team and Social Worker. These dyads showed symmetrical coping that enhanced short and long term well-being. A life course perspective emerges for individuals with heritable Rb. Mothers focused on their child's future health risk and Rb transmission to future generations. Fathers focused on possible socioeconomic disadvantage for their child. A disease-treatment matrix impacts the life course experience. Heritable Rb is referred to the single tertiary treatment centre in Canada. Regular travel from home and the absence of common social supports increases individual and dyadic stress for many affected parents. Implications for clinical practice include the importance of face-to-face meetings with clinicians as a primary parent coping strategy. Fathers should be actively encouraged to attend Rb appointments with their partner and child whenever possible. Those that did so enhanced both individual coping and positive dyadic coping outcomes. After clinician information, parents preferred brief, plain language pamphlets and brochures for take-away information. These were commonly lacking and internet resources were the default information source for parents. Some Rb parents gain substantial informal informational and social support from peer parents. Social media is the emerging channel among younger parents for that informal peer support.

Keywords

Stress, coping, stress process, life course, Retinoblastoma, fathers, role identity, discourses, dyads, dyadic stress, qualitative, interpretive description, family-centered, Pearlin, Elder, Cooper, Edley, Bodenmann.

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Introduction

Retinoblastoma (Rb) is a rare and complex genetic cancer presenting in one or both eyes and commonly diagnosed in infancy or very early childhood. Most childhood cancers share an important common feature: the treatment course is long, often intensive, and with uncertain outcomes. Retinoblastoma introduces new parents, individually and as dyads, to multiple acute stressors at diagnosis. Significant disruption of emerging social roles and transitions is common in both private and public spheres of life for these affected parents. Stressors can evolve to be chronic for individuals and dyads, sometimes impacting their life course trajectory.

Experiential father-focused research is largely absent in the child psychosocial oncology literature. This qualitative sociological research begins to bridge that knowledge gap by using in-depth, semi-structured interviews to examine stress and coping strategies and experiences of fathers as their child received care at specialized Canadian Rb treatment centres. The entire project emerged from a three-year research placement on the Retinoblastoma Team in the Department of Ophthalmology at the Hospital for Sick Children in Toronto. Multiple interdisciplinary Rb-related research projects, lengthy clinical observation, plus clear practice insights that emerged from ongoing supervised dialogue and interaction with clinical experts suggested that fathers holding different role identities may identify stressors differently, experience role strain differently, and cope differently than mothers encountering the same experience. Role-based stress and coping differences may have a distinct and separate impact on dyadic coping. Five research hypotheses emerged from this immersive research placement. The first three research hypotheses were grounded in the Stress Process Model (Pearlin, 1989) and assess fathers' individual stress and coping experiences. The Systemic-Transactional Dyadic Coping Model (Bodenmann, 2005) addresses dyadic stress and coping framed as hypothesis four. Hypothesis five addresses a broader intended research impact – providing clinical practice insights to improve coping and social support strategies for Rb parents. The life course process (Elder, 1994) layers across and informs aspects of all five hypotheses. Hypotheses are fully addressed in the Discussion (Chapter 6).

1. The relative absence of fathers from the medical domain during the early discovery and treatment phase results in increased personal and marital stressors;

2. Role strain persists for fathers as treatment outcomes for their child challenge traditional beliefs and activities expected from the father-child relationship;
3. Fathers experience more social isolation because they lack, or do not avail themselves of, available specific and general social supports;
4. Rb diagnosis and treatment disrupts the marital dyad at multiple levels, introducing role and relationship changes that are experienced as stressors;
5. Providing parents with easily understood information on their child's condition can mediate the experience of stress for both the parent and the dyad.

The medical context, relevant child psychosocial oncology research, and key sociological theories are presented in Chapter 1. The Interpretive Description methodology, along with a reflexivity statement and brief on the research ethics process, is presented in Chapter 2.

For this study, prior sociological research was adapted to create four functional fatherhood role categories for this study. Cooper's (2000) role identity work was amenable with this project because those categories embraced 'new economy' role definitions of fatherhood not commonly presented in the broader masculinities literature. Here, Cooper's original three categories are expanded to four – Traditional, Transitional-Transitional, Transitional-Superdad, and Superdad for analysis (see Chapter 1 section 'Using roles to define identity', for category details). Edley's (1991) fatherhood analysis provided three discourse dimensions – interpretive repertoires, ideological dilemmas and ambivalence, and subject positions – to contextualize fathers' narratives for cultural and personal references they used to construct their role identities (see Chapter 1 section 'Using discourse to define identity', for detail on dimensions). Integrating these two approaches provided a role-discourse classification matrix to categorize each father into one of four fatherhood roles based on narrative content at the time their child was diagnosed. The matrix is presented as Table 1 on page 20. Briefly, the four functional fatherhood roles are summarized as follows:

Traditionals: held an interpretive repertoire grounded in the idealized masculine role of public-sphere work and earning. Their own father was a primary reference, a subject position, for their fathering role identity. They expressed ambivalence around providing

caregiving and emotional support in the private sphere, deferring these functions to their partner.

Transitional-Traditionals: aspirations to the idealized masculine role represented by their own father were contested by the family caregiving impact of a nurturing partner. Dilemma and ambivalence around nurturing versus normative fathering roles in the family, from two distinct subject positions, was resolved through a form of role balance – active caregiving appended to the end of the regular work day.

Transitional-Superdads: aspirations of active caregiving contested a strong underlying commitment to work. Dilemma and ambivalence around active caregiving versus normative fathering roles was situationally negotiated with their partner, based on employer demands. Peer fathers were the primary subject position from which to assess successful attainment of work-life-family balance.

Superdads: ascribed to less-gendered, egalitarian role norms where family well-being extended into a multicultural, often multigenerational family system. Work role did not supersede active caregiving for these fathers. In health critical situations like Rb, these Superdads set aside the public-sphere for a primary role in private-sphere supportive care for their partner and child(ren). Dilemma and ambivalence focused on fairness/justice around impacts of cancer on the family and how to maintain the well-being of all members. Subject positions were less-gendered and need-based within the family system.

In Chapter 3, fatherhood role analysis was applied to stress and coping definitions in the Stress Process Model (Pearlin, 1989). Here, stress is an individual experience, dynamic and arising in the social world, and experienced in everyday social roles and social contexts. Psychosocial resources like mastery, coping, and social support are considered mediating or moderating resources. Direct or indirect effects may be physical or mental in nature, acute or chronic in impact. Similarities and differences in stress and coping by fatherhood role are, briefly: Spouses/partners were the primary mediator of stress for fathers across all four role categories. All types of fathers show affinity for problem-oriented coping while only Superdads engaged with emotion-oriented coping strategies (consistently a primary coping strategy for partners of all father categories). Traditional fathers regularly used social distraction or diversion in the public sphere as an effective avoidance/denial coping strategy.

They also actively resisted accepting instrumental supports from family and community sources, feeling this would negatively impact them as primary provider to the family. Superdads were unique in many aspects including spending considerable time with their partner on everyday activities. Facebook is emerging as a key mechanism for peer-to-peer informal social and informational supports for Rb families with active-treatment mothers the primary users.

Chapter 4 examines how each of the four fatherhood categories impacted dyadic stress and coping in Rb families. Bodenmann's (2005) Systemic-transactional Model of Dyadic Coping theorizes dyadic coping as distinct from individual coping (as in Chapter 3). The two commonly co-occur and separate analysis at the fatherhood-role level was required to disentangle and clarify father and dyadic coping strategies in the context of Rb treatment. Spousal support is key to dyadic coping and involves assessing process-level information like communication and intimacy difficulties, parental differences in child rearing practices, and the division of household and child-care tasks. Two primary impacts of effective positive dyadic coping are the reduction of stress for each partner and enhanced relationship quality. Negative dyadic coping was not uncommon in the period immediately following Rb diagnosis. Supportive Rb Team interventions, delivered in a family-centred model of care and often by a Social Worker, attenuated much of the negative coping related to shock, loss, and anger around diagnosis and social disruption of family life. This supported low-conflict, high-collaboration dyadic relationships and decision-making across all father types, at least during the critical treatment period. A feeling of "we-ness" – mutual trust, reliability, commitment, and the perception the relationship is supportive in difficult circumstances – for both partners resulted in symmetrical or complementary positive coping outcomes, in spite of high levels of acute stress. Superdad dyads, with fathers' regular and unique engagement in private-sphere and caregiving activities, experienced this reciprocal coping in everyday life before and after the Rb event. As treatment progressed and life returned to a new normal, Traditional and Transitional-Traditional dyads commonly returned to a range of positive and negative coping behaviours that were normative in their pre-Rb life. However, a permanent shift to positive, low-conflict, symmetrical coping was experienced by one Transitional-Traditional dyad and one Transitional-Superdad dyad over a period of years after Rb diagnosis. Delegated dyadic coping was a common occurrence in many families dealing with

heightened levels of caregiving during treatment. Mothers commonly decided to step out of the labour force to alleviate dyadic and family stress. Only Superdads actively used benefits for extended work leaves to focus on caregiving and spousal support in the private sphere.

Chapter 5 examines a life course perspective (Elder, 1994) related to bilateral Retinoblastoma. The life course perspective is directly affected by personal/family elements of biography, history, and social structure. With Rb, trajectory is also affected by a disease-treatment matrix that shapes the life course experience for fathers, dyads, survivors, and sometimes extended families. The bilateral, heritable form of the disease introduces treatment complexity, possible vision loss, and secondary risk that can shape life course trajectories for affected children, their future off-spring, and caregivers. Changing treatment modalities over the last 30 years has meant the younger Rb cohort no longer faces social disadvantages of facial disfigurement from radiation treatment. However, two clusters of cumulative disadvantage emerged in this study. First, a resource-based disadvantage occurred when lower family income was linked with Rb mothers taking extended (multi-year) leave from the work place for caregiving. Second, ease of access to health care services was reduced for families having to travel from northern and out of province locations for specialized Rb treatment. In spite of available external resources, strain was compounded for lower-income families. A disease-treatment matrix differentially streams patients for medical care and impacts family stress and coping. Unilaterals (children having Rb in one eye) are seen at secondary treatment centres closest to home. Treatment is simpler overall, and families are not subjected to prolonged social and relationship disruption. Complex unilaterals and all bilateral children (having Rb in both eyes) are treated in tertiary care in Toronto. Complex care is stressful for parents and child; those having to travel far or stay for prolonged periods of treatment experience social disruption and high levels of personal and dyadic stress. Distinct within-family and between-family linked lives perspectives emerged. Within-family linkages focused on understanding and mitigating risk associated with secondary non-Rb cancers of later adulthood and intergenerational transmission of Rb to future off-spring. Understanding genetics and access to ongoing counselling were important coping factors. Between-family linkages focused on peer mentoring for social and informational support. Participating bilateral families were the strongest advocates and openly wanted to ‘return the favour’ and mentor a new Rb family. Contextual treatment

information and informal social support from an experienced mentoring family were important coping factors. Finally, the life course perspective posits developmental change can occur at any point in time. Two fathers in this study permanently shifted from Traditional role identities to Transitional-Traditional and Transitional-Superdad role identities. These changes occurred over a period of years and with community-based professional counselling support.

Chapter 6 integrates research findings and discusses the manner in which fatherhood role impacts individual social stress and dyadic stress after a child is diagnosed with Retinoblastoma. Life course concerns differ between fathers and mothers. Mothers have a primary role in family health and focus on those life course implications for their child. Fathers' identity is primarily located in work and earning so their concerns relate more to loss of future socioeconomic opportunity for their child. Fathers strongly ascribed to one of the four role categories in everyday life. However, they could assume a caregiving-enhanced fatherhood role in the home for varying periods, conditional on critical family needs. For two of the fathers, the functional role transition seemed to fit with a psychosocial developmental model of permanent change occurring over several years. A proposed developmental model for permanent fatherhood role change is discussed including supportive psychosocial factors. This is followed by discussion of each of the five research hypotheses. The chapter concludes with implications to clinical practice and thoughts on future research.

Chapter 1

Medical, psychosocial, and sociological considerations

Retinoblastoma (Rb) is a rare genetic eye cancer that presents in one or both eyes. Almost half of affected children carry the heritable or germline predisposition to Rb through a mutation in both alleles¹ of the RB1 gene. The result is complexity in both medical condition and delivery of care. Retinoblastoma research has focused on medical treatment, genetic diagnosis, and counselling, with psychosocial factors explored from clinical and oncology perspectives. A true sociological perspective is absent from the research literature.

This chapter is structured in three parts. First is a concise review of the medical context – institutional and clinical delivery of care; referral and diagnosis; genetic analysis; treatment; follow-up, psychosocial care; and public awareness and education. Second is a summary review of psychosocial literature on childhood cancer showing the linkages to life course and stress process theory used in this study. Third is an overview of sociological theory applied in this research endeavor. Four sub-sections consider fatherhood role definitions (Cooper 2000; Edley 1991), a dyadic model of stress (Bodenmann, 2005), the life course perspective (Elder, 1974, 1994), and the stress process model (Pearlin, Lieberman, Menaghan, & Mullan, 1981) as each are integrated into this sociological research with Retinoblastoma parents.

Medical aspects

Retinoblastoma is a rare genetic eye cancer that commonly develops in late-stage fetal development or early infancy, with the large majority diagnosed before 3 years of age

¹ An allele is one of two variations of a specific gene, on one pair of chromosomes. One allele on one chromosome is inherited from each parent (National Retinoblastoma Strategy 2009, S45).

(Cross, 2010). From an epidemiology perspective, the incidence of Rb is 1 in 15,000 live births (Seregard, Lundell, Svedberg, & Kivela, 2004), with about 23 Canadian children newly diagnosed each year (National Retinoblastoma Strategy, 2009). Untreated Rb is fatal. Timely screening, diagnosis, referral, treatment, and follow-up delivered systematically by a multidisciplinary team results in cure for 98% of children with Rb (Douglas & Dockerty, 2005; Gatta et al., 2005; MacCarthy, Draper, Steliarova-Foucher, & Kingston, 2006). Many survivors retain useful vision (Gallie et al., 1996).

Many affected Canadian families do not receive optimal care. This is primarily due to inadequate knowledge of Rb in primary care settings and wide variations in access to treatment and follow-up (National Retinoblastoma Strategy, 2009). Retinoblastoma specialists are available in only a few centres of excellence, so a patient's province of residence significantly impacts access to timely, high-quality care. Delay in diagnosis and unequal access to genetic testing are currently the two primary inequalities that increase risk of poor outcomes. Most families are required to travel, some great distances, for treatment and after care. Not all provinces cover the costs associated with travel necessary to access care unavailable in the home province. This can place undue financial and emotional strain on many Rb families (National Retinoblastoma Strategy, 2009).

Institutional delivery of medical care in Canada

Since Rb is a rare disease, ongoing and new Canadian diagnoses must be managed by multidisciplinary teams with necessary resources and extensive ongoing experience in treating and managing these patients. Specialized Rb treatment centres are located in only a few major cities across Canada – Vancouver, Calgary, Toronto, Ottawa, Montreal, and Halifax – to provide optimal levels of care for this rare disease. Human and physical resources in secondary and tertiary Rb centres deemed necessary for optimal care of children with Rb and their families are summarized in Appendix A.

Primary Rb care occurs in the office of a family doctor, pediatrician, optometrist, or community-based ophthalmologist and a referral is made to a secondary Rb centre for

confirmatory diagnosis. A secondary Rb centre is a medical teaching hospital that has adequate resources and experience for primary treatment of unilateral Rb patients. Secondary centres refer bilateral Rb cases to tertiary centres, but participate in the care of bilateral cases after the initial treatment plan is developed by the tertiary centre. A tertiary Rb centre within a teaching hospital has all the characteristics mandatory in secondary centres, in addition to adequate resources and experience for treatment of bilateral² and rare trilateral³ Rb. The Hospital for Sick Children in Toronto is the tertiary centre in Canada with a multidisciplinary Rb team comprised of ophthalmologists, oncologists, social workers, nurses, imaging specialists, and child life professionals (National Retinoblastoma Strategy, 2009).

Seven dimensions of medical and care processes are used to describe the comprehensive nature of Rb treatment. Each describes key resources and services required for delivery of specific and complex treatment and follow-up in a timely and integrated manner. Level of care is based on disease complexity/severity and often requires travel to specialized treatment centres. In brief, these are:

Clinical delivery of Retinoblastoma care

Thorough vision screening in babies and young infants is recommended to start at 6 months to one year of age (Community Paediatrics Committee, 1998, 2009). This is optimal for surveillance and early detection of Rb as most cases present between 1 and 2 years of age (National Cancer Institute, 2016). Large-sample clinical case reviews indicated the most common presenting signs for Rb were leukocoria (white pupil), followed by strabismus (eye tracking) (Goddard, Kingston, & Hungerford, 1999; Abramson et al., 2003; Thorn, Gwiazda, Cruz, Bauer, & Held, 1994). Typically, parents

² Unilateral means only one eye is affected. Bilateral Rb means both eyes are affected by the disease and require active treatment (National Retinoblastoma Strategy 2009, S45).

³ Trilateral Rb patients have tumours in one or both eyes and a separate Rb tumour in the pituitary or suprasellar region of the brain (National Retinoblastoma Strategy 2009, S47).

bring their observations and concerns about the signs of possible Rb in their child to the attention of their primary caregivers. Leukocoria is the observed white reflection in the eye instead of the usual, healthy red-eye reflex (American Association for Pediatric Ophthalmology and Strabismus [AAPOS], 2016). This observation always warrants further examination, Rb being the most serious outcome. Photoleukocoria is evidence of leukocoria in a photograph produced with camera flash in low-light conditions. While not diagnostic of Rb, it has become much more prevalent in the clinical literature since the introduction of digital photography (Patel et al., 2012). Many parents notice the white reflex in family photographs and then seek medical attention. Today, parents often search the internet and identify signs they have noted in their child's eye to be consistent with Rb then bypass their primary healthcare provider and take their child directly to a hospital emergency room (National Retinoblastoma Strategy, 2009).

Referral and diagnosis

The primary healthcare practitioner is responsible for the referral to specialist care in a secondary or tertiary centre on a possible diagnosis of Rb. Recent research found 55 % of parents consulted more than one primary health care before obtaining a referral to an ophthalmologist (Goddard et al., 1999). Referral to a secondary or tertiary Rb centre is urgent and determined by geography, Rb expertise required and available, and the severity of the disease (National Retinoblastoma Strategy, 2009). Confirmatory diagnosis determines the extent of the disease and classifies the affected eye(s) based on distinct clinical features (Murphree, 2005). Unlike other cancers, pathology is not required. Confirmation of the clinical diagnosis is obtained only after the eye is enucleated (National Retinoblastoma Strategy, 2009).

Genetic analysis

Genetic testing⁴ improves early identification and supports the clinical requirement for early and aggressive tumour surveillance and therapy to optimize survival and vision outcomes (National Retinoblastoma Strategy, 2009). In addition, genetic counselling focuses on educating probands⁵, parents, and families about Rb heritability, future health risks and reproductive options for affected individuals (National Retinoblastoma Strategy 2009; Valenzuela, Chan, Héon, & Gallie, 2011). Heightened awareness is important for older adults having a heritable RB1⁶ germline mutation (Eng et al., 1993; Kleinerman et al., 2000; Fletcher et al., 2004). For these individuals, risk of secondary non-Rb cancer includes sarcomas, brain tumors, and melanoma (Fletcher et al., 2004) along with epithelial cancers of the lung, bladder, and breast (Marees et al., 2008; Yu et al., 2009). Total individual risk depends on multiple biological and treatment factors - underlying disease, genetic and family history, and prior exposure to chemotherapy and radiotherapy treatment (Garwicz et al., 2000). Finally, genetic testing can focus limited health care resources. In the absence of genetic status, current ophthalmology practice guidelines in Canada recommend children at risk (the proband and infant relatives) undergo up to 10 frequent, full-retinal examinations in the first 3 years of life (National Retinoblastoma Strategy 2009). When the RB1 status of the proband is known, the precise status of each

⁴ Genetic testing for mutation of the RB1 gene is key to predicting disease, reducing risks, and monitoring cancer progression. Retinoblastoma (Rb) is considered to be a “genetic” eye cancer, since almost 50% of affected children carry a heritable (germline), predisposing, RB1 mutation. Ninety percent of the RB1 germline mutations are new in the first affected child (the proband) and were not inherited from the parents. However, each child born to a person with a germline RB1 mutation has a 50% risk of inheriting the mutation. Although 90% of persons with a germline RB1 mutation will develop Rb, 10% will develop no tumours and will be unaffected carriers. However, their children will be at risk of developing Rb, even if the parent did not (Canadian Retinoblastoma Society, 2009, S17).

⁵ A proband is the first known person in a family affected by a heritable disease; used here to refer to the key family member in whom molecular studies seek the identity of the RB1 mutant allele (National Retinoblastoma Strategy, 2009, S47).

⁶ RB1 is a tumour-suppressor gene located on chromosome 13. Heritable Retinoblastoma can result when both gene copies are mutated in a cell (National Retinoblastoma Strategy 2009, S47).

relative and can limit surveillance to at-risk infants. Overall, short and long-term healthcare costs are lower.

Treatment

Retaining the affected eye, preserving vision, and producing good cosmetic results are also important but survival of the child is the overriding goal of treatment. Timely detection and treatment by multidisciplinary teams using current treatment protocols has resulted in successful treatment for more than 95% of patients (Gallie, Erraguntla, Heon, & Chan, 2004; Chan, Gallie, Munier, & Beck Popovic, 2005).

Establishing disease severity is a critical step in determining the most appropriate therapy for a particular eye and child, and to predict treatment outcome. The International Intraocular Retinoblastoma Classification (IIRC) was developed for prognosis staging of eyes with intraocular Rb (Murphree, 2005; Khetan, Chan, Wang, & Gallie, 2006). An initial treatment plan is developed after the first comprehensive examination under anaesthesia⁷ (EUA) and analysis of imaging and genetic testing results. Management of Rb takes into account vision status of both eyes (National Retinoblastoma Strategy, 2009). Final decisions are made in team discussions that include the parents (Panton et al., 2009; Community Paediatrics Committee, 2009). If there is parental disagreement with medical advice for enucleation, careful discussion includes compliance with life-saving therapy (Bowles et al., 2007). Cryotherapy⁸ is primarily used to lyse small peripheral tumours and for recurrences after chemotherapy (Hamel, Heon, Gallie, &

⁷ Examination Under Anesthesia is the standard treatment protocol for Rb. Patients undergo a general anesthetic and eye examination/treatment at regular intervals to assess and monitor progress (control) of the disease (National Retinoblastoma Strategy, 2009).

⁸ Cryotherapy - A trans-scleral cryoprobe cooled by nitrous oxide is used to double or triple freeze-thaw and destroy the tumour and underlying choroid. Ice crystals lyse the tumour cell membranes (National Retinoblastoma Strategy, 2009).

Budning, 2000; Abramson, Ellsworth, Grumbach, & Kitchin, 1985). Laser coagulation⁹ physically destroys viable tissue and tumour with heat and is used for small or residual tumours (Imhof, Moll, & Schouten-van Meeteren, 2001). Negative effects include scars that compromise vision, hemorrhages, tumour disruption and/or spread (Deegan, 2003; Schueler et al., 2003; Valenzuela et al., 2011). Enucleation is also performed when all other treatment modalities have failed (Gallie, Erraguntla, Heon, & Chan, 2004).

In the last decade, systemic chemotherapy has largely replaced radiotherapy as the primary treatment for Rb (Gallie et al., 1996; Chan, Gallie, Munier, & Beck Popovic, 2005; C. Shields, & J. Shields, 2002; Harbour, 2004). Chemotherapy is administered in pediatric oncology units at centres with special expertise in Rb (National Retinoblastoma Strategy, 2009). Systemic chemotherapy alone is rarely curative and requires consolidation of response by focal therapy^{7,8} (Gallie et al., 1996; Deegan, 2003). When chemotherapy/focal therapy fail, stereotactic radiation techniques deliver lower radiation dose to normal tissues surrounding the target area and reduce risk of secondary non-Rb malignancies or cosmetic deformities (Sahgal et al., 2006).

Follow-up

The last decade has seen tremendous advances in both the body of knowledge about childhood cancer and the technologies used to treat it. As a result, the population of childhood cancer survivors is growing. These individuals face potential chronic health issues that impact across the life course (Stevens, Mahler, & Parkes, 1998; Oeffiger et al., 2004; Hudson et al., 2003). Systematic follow-up of childhood cancer survivors offers positive benefits – monitoring for late effects of treatment and surveillance of ongoing health issues and secondary cancer. Health promotion, on-going counselling, and

⁹ Laser coagulation acts by physically destroying viable tissue and tumour with heat. Green, Red, and Infrared wavelengths are primarily for used for small tumours, residual tumour after chemotherapy, and recurrences after chemotherapy (National Retinoblastoma Strategy, 2009).

participation in research studies are also positive benefits for survivors and those providing health care (Ginsberg, Hobbie, Carlson, & Meadows, 2006).

Follow-up for Rb survivors has ophthalmology and oncology components. Active follow-up emphasizes ophthalmic surveillance for new and recurrent Rb tumours, visual rehabilitation, and monitoring of late treatment effects. Long-term follow-up focuses on survivorship – “monitoring adverse effects of cancer treatments, ongoing ophthalmological care, genetic counselling, and surveillance for secondary non-Rb malignancy” (National Retinoblastoma Strategy, 2009: S29). Oncology follow-up is provided to all children who have received adjuvant chemotherapy, radiotherapy, or stem cell transplantation to monitor for relapse of disease and late effects related to therapy (National Retinoblastoma Strategy, 2009).

Psychosocial care and access to services

Retinoblastoma emerges in infancy or very early in life and presents unique psychosocial challenges to families and patients, some of which impact families and affected individuals across the life course. At diagnosis, the burden falls on parents and close family members. Children are directly affected as they transition to school, then to adolescence and adulthood where they face many important life choices such as career, reproductive capacity, and risk to future offspring (Malkin & Friend, 1994; MacDonald & Lessick, 2000; Clarke, Rumsey, Collin, & Wyn-Williams, 2003; Rankin & Borah, 2003; Rumsey, Clarke, White, Wyn-Williams, & Garlick, 2004; Ulster & Antle, 2005). A wide range of other supports—school support, genetic counselling, visual rehabilitation, financial support, palliative care, and bereavement support (described below)—are important, but often not readily accessible to all families in need.

Family-centred health care acknowledges the vital role families play in a child’s well-being. This model of care promotes effective parental decision-making while respecting family and cultural values and beliefs. Mothers and fathers are encouraged to share and actively participate in caregiving during their child’s hospitalization. Family-centred care

depends on a comprehensive assessment including language and communication styles, family and cultural factors, psychosocial stressors, and health beliefs and practices (Institute for Patient and Family-Centered Care, 2016).

This model of care is delivered by interdisciplinary teams focused on attentive and interactive care to ensure adherence to treatment and optimal outcomes for the child (Ek, 2000; Hicks & Lavender, 2001). Treatment teams share this responsibility and provide parents easy and timely access to accurate information (Fazeli, Downie, Ulster, & Gallie, 2006). Research findings indicated Rb parents benefited greatly when structured psychosocial support was provided at diagnosis and integrated with medical treatment and follow-up (Ek, 2000). Effective parent-doctor communication is highly valued by parents and families and been shown to significantly decrease parental and family stress (Eden, Black, MacKinlay, & Emery, 1994; Shapiro, Perez, & Warden, 1998). Written information is often the best alternative to limited face-to-face clinician time. To be trustworthy, it must be accurate and appropriately written for the parents' language, culture, and level of education (Mitchell, Clarke, & Sloper, 2005). Alternatively, parents access many sources of information and social support.

Even with known risks on accuracy, online resources are highly utilized by many Rb families for education and to identify community resources. Informal peer social networks between Rb parents and Rb families are often established and maintained through email and social media tools and can be important sources of information and support. Parent/family days or weekends are an effective way of bringing affected individuals and families together with the goal of providing support and education, often with a recreational component (Hicks & Lavender, 2001). The Canadian Retinoblastoma Society provides this wide variety of formal and informal education and social supports for Rb families.

Lastly, effective palliative care must address psychosocial needs of the family, including sibling support, school intervention, spiritual support, anticipatory grief counselling, and bereavement follow-up. Community-based service is uncommon because few primary

care physicians or pediatricians are trained to provide palliative care to children (American Academy of Pediatrics, 2000). Home-based palliative care requires community-based nurses to act as continuous caregivers to the child and family and often extends to bereavement support (Hicks & Lavender, 2001; Martinson et al., 1978).

Public awareness and education

Increased awareness and education about Retinoblastoma, and especially presenting symptoms, may lead to earlier diagnosis and result in a higher likelihood of cure and better vision outcomes for affected individuals. This is important because parents, rather than healthcare professionals, are usually the first to notice the initial ocular signs of Rb. In one large study, the initial presenting sign in 80 percent of Rb cases was detected by a family member or friend (Abramson et al., 2003). Parents commonly use the internet to research a white pupil, and arrive at their first physician visit with a great deal of good information on leukocoria and its common causes.

Public awareness campaigns to educate the public on the signs and symptoms of leukocoria are likely to increase the rates of early detection of Rb and result in increased likelihood of parents taking their child to a specialist sooner. Awareness campaigns in developing countries have met with some success. In 2002, Brazil initiated a national campaign for early diagnosis of Rb (Tucca, 2013; Epelman et al., 2004). A public service announcement highlighting leukocoria as a symptom of eye cancer was broadcast on television stations and offered a toll-free telephone number the public could call to get more information. In addition, educational materials were provided to the general public, primary health care workers, and ophthalmologists. Similarly, in 2003, Honduras initiated an Rb awareness campaign promoting early diagnosis. Information was disseminated to parents via posters and flyers at health clinics, complemented by television and radio advertisements. In the years following this successful campaign, the percentage of Rb patients presenting in the clinic with advanced (fatal) extraocular disease was 35 percent as compared to 75 percent in the 8 years preceding the campaign (Leander et al., 2007). Public awareness campaigns have not been successful in Canada.

Health system treatment pathways

The Canadian health care system has a formal pathway model for Rb referral and treatment. Primary care responsibility resides with a family doctor, pediatrician, optometrist, or community-based ophthalmologist. A referral to a secondary Rb centre is made at the first clinical concern of Rb. These secondary centres may be thought of as regional referral and treatment centres and are often the closest referral point for the affected child and family. After referral of a potential diagnosis of Rb, the secondary Rb centre will confirm the diagnosis and develop the initial treatment plan. Case consultation often occurs with a tertiary Rb centre (National Retinoblastoma Strategy, 2009).

Secondary Rb centres are medical teaching hospitals with adequate resources and trained personnel to diagnose unilateral and bilateral Rb, treat unilateral Rb cases, and provide follow-up care for complex Rb cases initially treated at a tertiary centre. Secondary centres may participate in the care of bilateral cases in consultation with the tertiary Rb centre. A social worker specifically assigned to Rb families is recommended at secondary centres, especially when there will be shared management and follow-up of bilateral cases (See Appendix A).

A tertiary Rb centre within a teaching hospital has the mandatory characteristics of a secondary centre, plus adequate resources and trained personnel to treat bilateral and rare trilateral Rb patients. Tertiary centres manage Rb patients with a dedicated multidisciplinary Rb team that includes ophthalmologists, oncologists, social workers, nurses, imaging and child life specialists (National Retinoblastoma Strategy, 2009).

Psychosocial research in childhood cancer

Cancer is often thought of as an adult disease. What is not appreciated is the fact that the highest rates of childhood cancer occur in the first year of a child's life, when parents are in the early stages of their family life cycle (Van Dongen-Melman, 2000; McGrath, 2001). The majority of psychosocial research in pediatric cancer relates to childhood

leukemia. A relatively common childhood cancer, leukemia provides an ample pool of subjects for multiple and on-going research studies (McGrath, 2001; Woodgate, 1999, 2000; Sloper, 2000; Stuber et al., 1994; Murray, 2001).

Research literature on the psychosocial impacts of Rb is rare, like the disease itself. An extensive literature review showed only four papers (Ulster & Antle, 2005; Fears & Parry, 1995; Rumsey et al., 2004; Ek, 2000). Two studies explored emotional responses experienced by Rb patients and family members over the course of diagnosis, treatment, and short-term follow-up (Rumsey et al., 2004; Ek, 2000). One was a qualitative case study that examined immediate and short-term patient and parent experience with bilateral enucleation and blindness (Ulster & Antle, 2005). Three of the four studies incorporated quality of life measures as a way to assess longer term or life course impacts of the disease on affected children and their families over the longer term (Ulster & Antler, 2005; Ek, 2000; Sheppard, Eiser, & Kingston, 2005).

Considered as a whole, this body of research literature indicated young children diagnosed with cancer, as well as their parents, experience distinct psychological stress with a wide range of exposure from the initial crisis, through active treatment and short-term follow-up, to long-term follow-up into adulthood (Ek, 2000; Sheppard et al., 2005; Boman, Lindahl, & Bjork, 2003). Common emotional distress symptoms experienced by parents of children with cancer include uncertainty, worry, sleep disturbances, anxiety, somatization and depression (Tuttle, 1984; Eiser, 1997; Van Dongen-Melman, 2003). Parents reported the highest levels of stress in the first two months after their child is diagnosed with cancer (Santacroce, 2002; Lahtenmahki, Huostila, Hinkka, & Salmi, 2002). Uncertainty caused anxiety for many parents (McGrath, 2001; Van Dongen-Melman, 2000; Sloper, 2000; Mastroiannopoulou, Stallard, Lewis, & Lenton, 1997) and that stressor remained long after treatment was complete and the child considered cured (Ulster & Antle, 2005; Byrne, Fears, Whitney, & Parry, 1995). Lifestyle changes due to increased caregiving load had significant financial implications for parents, often

resulting in changing terms of employment and increased expenses (Sloper, 2000; Yantzi, Rosenberg, Burke, & Harrison, 2001; Byrne et al., 1995).

Interventions to moderate impact of stress

Active parent support programs positively impact the long-term well-being of the child and the family. These structured interventions reduce parental anxiety and improve both parental decision-making and treatment compliance (McGrath, 2001; Mastroiannopoulou et al., 1997; Eden et al., 1994). Many parents identified access to disease and treatment-related information to be key to effective support (Eden et al., 1994; Sloper, 1996).

Interdisciplinary healthcare teams are an effective way to meet the diverse information and psychosocial needs of childhood cancer families (Mitchell, Clarke, & Sloper, 2005).

This researcher coauthored research identifying a cluster of parental risk factors that impact the understanding of complex medical information that may in turn influence the informed consent process. Younger parents were at greater risk of not fully understanding treatment complexity, even with high levels of education. English language fluency significantly affected parental understanding of Rb treatment and risk. Physician language fluency may also impact effective communication with parents, suggesting the importance of readily available language translation services whenever parent-physician communication involves informed consent. (Panton et al., 2009). Presenting a visual timeline of treatment allowed parents with lower levels of education attainment or weaker language skills to quickly and accurately understand the complexity and risk of treatment, potentially offsetting the disadvantage of lower education and language barriers (Panton et al., 2009; Chiu, Dimaras, Downie, & Gallie, 2015).

Life course and stress process in childhood cancer

A diagnosis of cancer is a traumatic event for young children and their families (Stuber et al., 1994; Sloper, 2000; McGrath, 2001; Van Dongen-Melman, 2000). The event introduces a unique set of circumstances requiring individual and family adaptation during the treatment period and beyond (Ulster & Antle, 2005; Shapiro et al., 1998; Stovall, 1993). Cancers of infancy or very early in life place the psychological and

caregiving burden on parents and close family members (McGrath, 2001; Van Dongen-Melman, 2000; Shapiro et al., 1998; Boman et al., 2003). Acute and chronic strain emerges from sudden and unexpected parental and caregiving role changes, increased care-giving demands, and the threat of cancer recurrence (Ulster & Antle, 2005; Rumsey et al., 2004; Ek 2000; Sheppard et al., 2005; Boman et al., 2003; Murray, 2001). Many parents with a cancer-affected child experience levels of stress and social disruption similar to parents of children with physical or developmental disabilities (Dyson, 1997; Hancock, Wilgosh, & McDonald, 1990; Leyser, Heinze, & Kapperman, 1996; Leyser & Hienze, 2001; Troster, 2001; Sloper, 1996; Eden et al., 1994; Bessell, 2001; Mitchell, Clarke, & Sloper, 2005; Downie & Gallie, 2005). These stressors may force adjustments in family roles and organization (Mitchell, Clarke, & Sloper, 2005; Yantzi et al., 2001). Medical phobias can develop during or after treatment and may result in avoidance behaviours over the longer term, possibly having negative health consequences (Ulster & Antle, 2005; Ek, 2000).

During the school years, parents often reported teasing or bullying if cancer treatment resulted in changes to physical appearance (Rumsey et al., 2004; Ek, 2000; Sheppard et al., 2005; Mastroiannopoulou et al., 1997). Teasing and bullying about physical appearance during childhood and adolescence was associated with degrees of poor self-esteem and social anxiety in adulthood. Long-term implications are apparent in various life domains, including education attainment, income attainment, social integration, and employment outcomes (Rumsey et al., 2004; Byrne et al., 1995; Woodgate, 2000; Ek, 2000; Ulster & Antle, 2005; Eiser, 1997). The transition to adulthood introduces other challenges as the affected individual begins to understand their medical history, the implications of the disease and treatment, and potential risks carried into adulthood (Ulster & Antle, 2005; Malkin & Friend, 1994; MacDonald & Lessick, 2000; Clarke et al., 2003; Rankin & Borah 2003; Rumsey et al., 2004). Generational considerations emerge, centered on the risk to future offspring (Tuttle, 2004). Ethical, emotional, and psychological issues pertaining to genetic testing are brought into any decision matrix (National Retinoblastoma Strategy, 2009).

Sociological theory

Both child and family are exposed to a great deal of social disruption and emotional stress throughout the treatment course for most childhood cancers (Mitchell, Clarke, & Sloper, 2005; Stuber, Meeske, Gonzalez, Houskamp, & Pynoos, 1994). Limited sociological research indicated social support was a protective factor that mediated negative effects of stressful life events and reduced burden of care (Kim, Duberstein, Sorenson, & Larson, 2005). Social support measures have been operationalized in many different ways (Secco & Moffat, 1994) but the overall direction of the research indicated attenuating effects of social support was related to perceived social support and not size of social network providing the support (Speechley & Noh, 1992; Avison et al., 1993).

Constructing fatherhood role identity

Fathers and fatherhood research has received expanded attention in social research, policy development, and cultural media in recent years. This new focus reflects a number of diverse agents promoting the active involvement of fathers in family life as something that is good both for children and for men themselves (Robb, 2004). Until recently, two key research domains have been: (i) associating child developmental outcomes with patterns of father involvement; and (ii) investigating how fathers balance economic work, household work, and caregiving work (Marks & Palkovitz, 2004).

Connell's foundational scholarship on the definition and practice of masculinity examined the discursive construction of men's identities (Brod & Kaufman, 1994; Mac An Ghail, 1996). Connell and colleagues suggested masculine identity was plural and dynamic, having a range of dominant to subordinate definitions (Connell, 1987, 1995, 2003; Kimmel, Hearn, & Connell, 2004; White, 1994; Gregory & Milner, 2011; Brod, 1987; Brod & Kaufman, 1994; Kimmel, 1987; Brandth & Kvande, 1998; Marsiglio, 1998). These plural masculinities are linked to each other (Edley & Wetherell, 1997) and conflict can arise between these definitions (Cooper, 2000).

The majority of fatherhood research has developed general typologies while minimizing inter- or intra-individual variability in styles and commitment. Marks & Palkovitz (2004) argued that “categorical typing helps establish parameters for research; identifies what researchers should pay attention or sensitize to; and organizes complexities of contemporary individual and family life.” (p. 114). To be sure, fathers will offer unique histories and differ in developmental trajectories, styles of interaction, and levels of involvement with their family. However, there is legitimacy to carefully generalized discussions of fatherhood because most fathers share some universal characteristics (Marks & Palkovitz, 2004; Lamb, 2000; Pleck, 1987).

In a variety of ways, fatherhood is socially (White, 1994; Brandth & Kvande, 1998; Gregory & Milner, 2011), culturally (Marsiglio, 1998), and discursively constructed (Robb, 2004; Edley, 2001). Comparatively little work has explored men’s experience as fathers and how personal meanings intersect with public discourses (Robb, 2004). Often described in terms of fixed roles or behavioural sets, fathering styles are varied and fluid, determined by multiple factors that are dynamic at both the individual and cultural level (Horna & Lupri, 1987). Movement or development through diverse styles of fathering may be more the rule than the exception (Marks & Palkovitz, 2004).

Using discourse to define identity

Edley (2001) approached understanding the meanings fatherhood holds for men by paying attention to how they talk about their experience as fathers, then analyzed the frameworks and strategies they used to make sense of their experience. He used three distinct concepts for analysis – interpretive repertoires, ideological dilemmas, and subject positions.

Interpretive repertoires

Interpretive repertoires can be described as “culturally-available frameworks that enable individuals to make sense of their experience” (Robb 2004, p. 123; Potter & Wetherell, 1987). In a given research context, participants will draw on a number of

shared meaning-making frameworks to describe their fathering experiences (Robb, 2004). Edley argued interpretative repertoires held advantages over discourses. Interpretive repertoires were less monolithic, smaller and more fragmented, as compared to discourses. They “offered speakers a whole range of different rhetorical opportunities” (Edley, 2001, p. 202) to uniquely describe and differentiate individual fatherhood experiences.

Ideological dilemmas

Ideological dilemmas capture the fragmentary and contradictory nature of everyday experiences. These lived ideologies are “inherently dilemmatic, or structured around contradictions” (Robb, 2004, p. 125). Exuding ambivalence, these everyday identity experiences revolve around sets of oppositions that have to be worked through and resolved (Billig et al., 1988).

Subject positions

Subject positions involve individuals locating themselves with respect to both external discourses and real or imagined others (Robb, 2004) in order to “... view themselves and make sense of their experience” (Edley, 2001, p. 209). In negotiating personal identities, fathers frequently defined themselves by comparing and contrasting themselves with others. Sometimes this was their spouse or partner, more often it was other men, with the most significant other man being their own father (Robb, 2004).

Using role to define identity

Cooper (2000) identified three primary fatherhood identities contextualized by modern definitions of masculinity – “Traditionals”, “Superdads”, and “Transitionals” (p. 391-394). In Cooper’s research, fathers were knowledge workers experiencing normative workplace controls that often kept them focused on the work life rather than family life. This embedded “masculine ethic” (Kanter, 1977, p. 22) often resulted in role strain narratives from fathers and conflicted life experiences in family relations.

Traditional role

Traditionals see the domestic division of labor along traditionally gendered lines. They emphasize their work and the income it provides while having a curtailed definition of care and the inability to sympathize with their spouse. This exhibits as inattention to the needs of others and leaving the family caregiving role to their wife or partner.

Traditionals were also the least likely to access paternity leave for caregiving. Traditional fathers “commonly relinquished or shifted caring work to their spouse and often encountered resistance” (Cooper, 2000, p. 392) from their spouse in return.

Superdad role

Superdads sacrifice their personal selves in order to invest heavily in both career and family obligations. They hold an egalitarian gender ideology whether or not their wives stay home. Superdads hold a care orientation that engenders strong emotional connections to family members. For them, caregiving includes attentiveness to emotional, physical, and spiritual needs of those around them. Caregiving includes emotion work, care work, and paid work. Holding this broader definition of care enables them to anticipate needs of family members and empathize with their partners (Cooper, 2000, p. 391).

Transitional role

Transitionals exhibit qualities of both Traditionals and Superdads. Paradox is present: like Traditionals, they may renege on their egalitarian ideology to some degree and leave much of the caregiving work to their wives; like Superdads, they want to be involved fathers and take responsibility for some of family work. Some lean towards being a Superdad, others towards a Traditional role. Transitionals have a harder time balancing work and family commitments. Their caregiving orientation is held more loosely so they have fewer problems handing off emotion work and frequent caregiving duties to their spouse (Cooper, 2000, p. 393).

Linking role and discourse for analysis

Prior research suggested fathers' discourse could be linked to role identity. Rutherford's (1988) perspective offered a very good comparison of two dominant masculine discourses related to fatherhood. He labeled these subject positions as 'retributive man' and 'new man'. Retributive man is akin to Cooper's 'Traditional' father and represents a traditional form of masculinity, archetypal in nature and aligned to culturally dominant hegemonic forms described by authors like Connell (1987, 1995). Here, the father was the major breadwinner in the family and the principle source of authority for the family. "He is tough, competitive and emotionally inarticulate" (Edley & Wetherell, 1997, p. 204). New man is most closely aligned with Cooper's 'Superdad' definition of fatherhood. "He is the ideal partner for the modern, liberated, heterosexual woman ... a softer, more sensitive and caring individual, who also avoids sexist language, changes nappies, and loves to shop for his own clothes." (Edley & Wetherell, 1997, p. 204). Cooper's 'Transitional' definition of fatherhood clearly aligns to a central ground, that area of contested masculinity/fatherhood present in modern Western societies. This conflicted role identification is experienced and expressed by these fathers in different ways and across multiple life contexts.

Cooper's three role categories were expanded to four for this research. The much larger sample size permitted a descriptive or functional split of the Transitional category. See Chapter 3, 'Fatherhood roles' section for detailed description of Traditional, Transitional-Transitional, Transitional-Superdad, and Superdad categories used in this study. Edley's (1991) fatherhood discourse analysis provided three dimensions – interpretive repertoires, ideological dilemmas and ambivalence, and subject positions – to contextualize fathers' narratives for cultural and personal reference points in categorizing their role identities. Integrating these two approaches provided a role-discourse classification matrix to categorize each father into one of four fatherhood roles based on specific narrative content provided in interviews. Table 1 (following) differentiates key discourse elements by each of the four role categories.

Table 1: Fatherhood role-discourse matrix

| Cooper-Role → Edley -Discourse ↓ | Traditional | Transitional- Traditional | Transitional- Superdad | Superdad |
|---|--|--|--|--|
| Interpretive repertoire | Idealized masculine role in public sphere Private-sphere role minimized; caregiving deferred to partner | Aspirational to idealized masculine role Regular but limited caregiving appended to end of work day | Aspirational caregiving role & commitment to traditional work role Active caregiving role negotiated with partner but around employer needs | Egalitarian; well-being of extended family members; how to support all needs Private-sphere caregiving equally important to public-sphere work; caregiving was primary role in crisis |
| Ideological dilemmas and ambivalence | Emotional expression & active caregiving role | Contested role: Nurturing partner vs. normative father Work-care role balance | Contested role: Active caregiving vs. primary work commitment Work-life-family balance | Issues of fairness and social justice (cancer-related in this study) |
| Subject positions | Cultural ideals from own father | Boyhood ideals from own father; competing role of nurturing partner | Peer fathers | Less-gendered, need-based roles based in multicultural, multigenerational family system |

Dyadic stress model

A considerable body of psychosocial research has examined marital stress in the context of families with a seriously ill child, including cancer. Reported findings have varied widely across time and by study design. Early findings indicated higher levels of marital distress and higher divorce rates for parents raising a child with a serious illness (Tew, Payne, & Laurence, 1974). More generally, there are conflicting findings on marital strain and outcomes with families of seriously ill children (Quittner et al., 1998; Walker, Manion, Cloutier, & Johnson, 1992; Barbarin, Hughes, & Chesler, 1985; Holmbeck, et

al., 1997). Depending on methods and measures, psychosocial researchers examining family outcomes across a variety of serious childhood illnesses have suggested higher overall stress in the family system (McCubbin, Cauble, & Patterson, 1982) as well as higher stress for individual caregiving parents (Holmbeck et al., 1997; Thompson & Gustafson, 1996). Other pediatric psychosocial research has indicated the majority of families with chronically ill children demonstrated healthy adaptation in spite of the increased demands and strains (Kazak, 1997; Quittner & DiGirolamo, 1998; McCubbin, 1989; Kazak & Nachman, 1991; Patterson & Garwick, 1994). The handful of studies that included fathers showed substantive differences: fathers experienced heightened stressors related to finances and emotional attachment to the child while mothers have reported greater stress in managing daily routines and medical regimens (Quittner, DiGirolamo, Michel, & Eigen, 1992).

Attempting a comprehensive examination of these questions, Holmbeck, Li, Schurman, Friedman & Coakley (2002b) used multiple data sources and methods to examine family stress when caregiving for a seriously ill child. Triangulated results showed disruption from the child's illness did not translate into higher levels of family conflict and stress, confirming some of the previous research findings (e.g., Kazak, 1997; Quittner & DiGirolamo, 1998; McCubbin, 1989; Kazak & Nachman, 1991; Patterson & Garwick, 1994). However, these parents experienced heightened individual stress with additional negative life events such as low income providing cumulative stressors, also confirming prior research (e.g., Goldberg, Morris, Simmons, Fowler & Levison, 1990; Kazak & Nachman, 1991; Holden, Chmielewski, Nelson, & Kager, 1997).

Measurement issues were identified as a key reason for these inconsistent findings in the literature, related to marital stress with chronically ill children (Holmbeck et al., 2002b). Some studies focused on stressful life events rather than specific tasks and strains of caregiving; many used non-categorical approaches that combined diverse medical conditions, wide age ranges, and different phases of the illness into one sample (Varni, Wilcox, & Hanson, 1988). At the same time, studies ignored important considerations

like severity and duration of the stressor, life course stage of child and family, and the primary social roles affected by the strain (e.g., parent or spouse; Pearlin et al., 1981; Quittner, 1992; Rolland, 1987). As a result, weak relationships between stress and coping have been generally reported across the research literature (Wallander et al., 1989; Wolf, Noh, Fisman, & Speechley, 1989), with little discovery of processes underlying successful coping (Drotar, 1997).

The diversity of findings in this area underscore the need to replace global and individual measures of stress and coping. Researchers should examine dyad-specific sources of strain – communication and intimacy difficulties, parental differences in child rearing practices, and the division of household and child-care tasks (Bristol, Gallagher & Schopler, 1988; Klinnert, Gavin, Wamboldt, & Mrazek, 1992). Dyadic process-level information is essential for a deeper understanding of stress and coping in families (Holmbeck, Coakley, Hommeyer, Shapera & Westhoven, 2002a; Wallander et al., 1989; Kazak, 1997). Information needs to be collected from both parents (Holmbeck et al., 2002a; Holmbeck et al., 2002b) in a systems context (Bodenmann, Pihet, & Kayser, 2006) to deepen the contextual understanding of how parents and families adapt and are transformed by these critical life experiences (Costigan, Floyd, Harter & McClintock, 1997).

Early, individual-effects models of dyadic stress

When social scientists began to study coping with stress in the 1960s and 1970s, coping was considered an individual phenomenon (Lazarus & Folkman, 1984; Pearlin & Schooler, 1978; Pearlin, 1989; Pearlin, Mullan, Semple, & Skaff, 1990; Pearlin & Turner, 1987). The transactional concept of stress and coping (Lazarus & Folkman, 1984) was the most recognized and applied stress paradigm in social psychology at that time. The stress process (Pearlin, 1989) emerged as the dominant sociological paradigm.

Pearlin and colleagues authored several foundational articles detailing a comprehensive model of caregiver stress and role strain (Pearlin, 1989; Pearlin, Mullan, Semple, &

Skaff, 1990). Conceptually, this model integrated four components of role strain relevant to marital stress: (i) parenting role stressors (includes caregiving activities); (ii) frustration of role expectations (expectations and disappointments about the division of labor); (iii) parental role conflict; and (iv) affective exchange (giving and receiving affection; Pearlin & Turner, 1987). Still, measurement was at the individual level and dyadic stress extended from the individual experience. Individual coping scores of each partner were compared to produce congruency or discrepancy rankings (Bodenmann, Pihet, & Kayser, 2006). That body of literature has examined constructs related to stress (Quittner & DiGirolamo, 1998; Wallander et. al. 1989) as well as conflict and cohesion (Cox & Brooks-Gunn, 1999) in marital dyads and family systems.

In the early 1990s, researchers began to emphasize the significance of the social context and the role of significant others in managing stressful encounters (Ledermann et al., 2010). A genuine definition of couples' coping began to emerge at that time. Several integrative dyadic stress and coping models have been developed: empathic coping (DeLongis & O'Brien, 1990), relationship-focused coping (Coyne & Smith, 1991), and systemic-transactional dyadic coping (Bodenmann, 2005). All three approaches extend the individually-oriented transactional view of stress to a dyadic process where the coping reactions of one partner take into account the stress signals of the other partner.

In each of these three approaches, dyadic coping is a novel construct clearly distinguished from social support. Three main differences between social support and dyadic coping were identified as: i) support occurring as part of dyadic coping is spousal support which is distinct from support provided by other persons (friends, kin, neighbors, etc.). A spouse is often the primary and most important source of support in stressful situations so spousal support carries a different meaning than social support from others; ii) unlike social support, dyadic coping involves both partners to ensure each partner's satisfaction and wellbeing, which in turn ensures individual well-being and the smooth functioning of the dyadic relationship; and iii) dyadic coping also includes other types of stress

management behaviors that both partners actively engage when faced with common stressors (Bodenmann, Pihet, & Kayser, 2006).

A systemic-transactional dyadic coping model

Coyne and Smith (1991) identified active engagement and protective buffering as two forms of dyadic coping. In active engagement, one partner attempts to involve the other by discussing the stressful situation with their partner, exploring his or her emotions around the situation, in an attempt to constructively solve problems. Protective buffering emotionally relieves the partner through hiding or yielding to concerns, minimizing worries, suppressing anger, and protecting one's partner from upset and burden.

Building on this early conceptualization, Bodenmann (1995, 1997, 2005) developed a comprehensive model of dyadic coping in close relationships, a systemic-transactional perspective derived from observation of interactions between spouses under stress. Two types of stress are possible in this model: direct or relationship stress and indirect or external stress (Bodenmann, Ledermann, & Bradbury, 2007; Randall & Bodenmann, 2009). This theoretical framework for systemic-transactional dyadic coping is based on two key assumptions: First, stress and coping represented a dyadic (systemic) phenomenon. Second, dyadic coping with stress included both stress expression and dyadic support (Bodenmann, 2005; Ledermann, et al., 2010). Dyadic stress and dyadic coping are viewed as parts of an interpersonal process involving both partners (Bodenmann, 2005). Dyadic stress is a specific stressful encounter that directly or indirectly affects both partners and triggers the coping efforts of both partners within a defined time frame and geographic location – "... couples are embedded in a shared social context and dyadic coping assumes their interdependence, common concerns, and mutual goals and stimulates joint problem-solving and common emotion-focused coping activities" (Bodenmann, 2005, p. 33). All dyadic coping occurs in addition to individual coping (Bodenmann, 1995, 1997).

Dyadic stress can affect both partners simultaneously or sequentially. Direct or relationship stressors originate within the relationship and impact the marital relationship (e.g., differing attitudes or goals; shared problems; or marital conflict). Direct dyadic stress occurs when both partners face a common stressor at the same time (e.g., child's Rb diagnosis in this research context; Bodenmann, 2005). Indirect or external stressors originate outside the couple and involve interpersonal interactions in the social environment – co-workers, neighbours, or extended families. Handled adequately, these stressors never impact the partner. Unresolved stressors, like workplace stress, are likely to migrate into the home and marital relationship to cause indirect dyadic stress. Stressor timing very is important to differentiate between individual and dyadic stressors.

The two primary impacts of effective dyadic coping are reduction of stress for each partner and enhanced relationship quality. In this model, one individual's well-being depends on the well-being of their spouse and his/her integration in the social environment. Effective dyadic coping should help both partners manage their stress. Positive dyadic coping has a second distinct effect, fostering a feeling of “we-ness” – meaning mutual trust, reliability, commitment, and the perception the relationship is supportive in difficult circumstances (Bodenmann, 2005, p. 41).

The life course perspective

Yet unrecognized life course tenets were applied in early twentieth century research examining adversities faced by recent Polish immigrants to America (Elder, 1985; Elder et al., 2003). In 1959, C. Wright Mills proposed a new orienting concept for behavioural sciences as “... the study of biography, of history, and of the problems of their intersection with social structure.” (p. 149). However, the life course perspective was not developed as a methodological approach in sociology until the 1960s (Elder, 1985). Rapid social change in America – the Vietnam War and the Civil Rights Movement – combined with economic volatility focused researchers on the interplay between social change and the life course trajectories of individuals (Elder et al., 2003). Longitudinal studies during that period, combined with ongoing sociological research, began linking

social change and life patterns of individuals (Giele & Elder, 1998; Elder et al., 2003; Elder & Giele, 2009; Heinz, Huinink, Swader, & Weymann, 2009).

This interdisciplinary sociological framework emerged from three distinct developmental research traditions – life-span development, social roles and relationships, and age and temporality (Elder, Shanahan, & Jennings, 2015). Glen Elder (1985, p. 5) described the life course as “the interweave of age-graded trajectories, such as work careers and family pathways, that are subject to changing conditions and future options, and to short term transitions ranging from leaving school to retirement”. It embraces a social constructionist view of lived experience (Elder, 1994; see also Bandura, 1977; Goffman, 1978; Searle, 1995; Berger & Luckman, 1991), integrating historical and current social contexts, personal biography, agentic factors, and intergenerational influences that lead to unique developmental pathways (Elder, 1974, 1995, 1998, 1994; Turner, Pearlin, & Mullan, 1998; Elder, Johnson, & Crosnoe, 2003; Settersten, 2003; Hunt, 2005; Macmillan & Copher, 2005). Life course analysis is sensitive to the impact of early transitions on later experiences and how early experiences influence outcomes in later life (Clausen, 1993; Elder, 1994, 1998). In sum, a life course perspective describes unique patterns of social change and, at the same time, accounts for person-level differences in strengths, vulnerabilities, opportunities, and challenges (Elder, Shanahan, & Jennings, 2015).

Central principles

There are five central principles in the life course perspective – Lives and historical times; Timing of lives; Linked lives; Human agency; and Life-span development (development and aging as a life-long process) (Elder, 1994; Giele & Elder, 1998). The life-span principle is a key linking mechanism in the life course perspective. It highlights human development and aging as life-long processes and the importance of investigating impacts of both childhood conditions across the life course and agentic acts in adulthood (Mortimer & Shanahan, 2004). This dynamic creates unique patterns of social change and accounts for person-level differences in strengths, vulnerabilities, opportunities, and

challenges (Elder et al., 2015). In essence, individuals are produced, sustained, and changed by their social context (Gottlieb, Wahlsten, & Lickliter, 2006).

Lives and historical times

This principle suggests “the life course of individuals is embedded in and shaped by the historical times and places they experience over their life time.” (Elder, 1998, p. 3).

Restated: an individual’s life course will be simultaneously influenced by the historical context and geographic factors (Elder et al., 2003). Historical effects may present as a cohort effect, where social change differentiates life patterns of successive cohorts.

Historical effects may also present as a period effect, where change is quite uniform across successive birth cohorts. Individual outcomes ultimately depend on what the individual brings to the change process as well as the nature and severity of the change event (Elder, 1994; Kuh & Hardy, 2002).

Timing of lives

“The developmental impact of a succession of life transitions or events is contingent on when they occur in a person's life.” (Elder, 1998, p. 3). This principle incorporates both social meaning and social timing concepts into life events and transitions. Social meaning applies to structural criteria like age that brings a temporal or age-graded perspective to both social roles and life events. Social timing refers to the incidence, sequencing, and timing of roles, events, and transitions. Social timing is also relevant to the scheduling of multiple trajectories and their synchronicity with norms, indicating the importance of life stage points for social change (Elder, 1994; George, 2009). Timing of lives posits both causes and consequences of transitions and events are to a certain degree dependent on when they occur in the life course trajectory (Mortimer and Shanahan, 2004). As well, ongoing events will affect respective cohorts differently (George, 1993).

Linked lives

Elder (1998) wrote that “lives are lived interdependently, and social and historical influences are expressed through this network of shared relationships” (p. 4). Individual

social worlds dynamically interact with those of family, friends, and co-workers across the life span. Social support and social regulation emerge from these relationships, often described in terms of socialization, behaviours, and generational attributes (Elder, 1994). Individual experience of macro-historical change to a great extent emerges from dynamic interactions of these multiple social worlds (Elder & O’Rand, 1995).

Human agency

“Individuals construct their own life course through the choices and actions they take within the opportunities and constraints of history and social circumstances.” (Elder 1998, p. 4; see also Elder 1994; Clausen 1993; Giddens 1984; Sewell 1992 for more on agency). Restated, individual social trajectories shape developmental processes and outcomes. The relationship is reciprocal with developmental processes affecting social trajectories (Elder, 1994). Understanding these selection processes is important in life course research: individual differences produce diverse behaviours and outcomes when interacting with changing environments (Elder, 1994; Elder & O’Rand, 1995). Early choices are especially important to the understanding of later outcomes (Elder, 1994).

Life-span development

From a scientific perspective, the study of human development was primarily devoted to child development through the first decades of the 20th century, expanding to adolescent development. In the latter half of the 20th century developmental research began to focus on adult development and aging – life-span development (Settersten, 2003; Alwin, 2012). In this early practice, the life span was represented as a sequence of developmental stages and transitions, each an age-specific domain for specialized study – e.g., child, adolescent, or elderly (Elder & Shanahan, 2006).

From a biographical perspective, life span represents the length of an individual’s life and signals the temporal scope of inquiry (Elder, 2000). This perspective suggests “developmental change occurs over the entire course of life; it is synonymous with aging in the broadest sense. Aging is not limited to any particular time of life; neither is

development” (Featherman, 1983, p. 2). Human development means within-person change resulting from numerous biological, psychological, and social factors (Alwin, 2012). Life-span development describes multidimensional and multidirectional processes of growth or change or aging that involve both gains and losses. The uniqueness of individual biographies and the diversity of life patterns flow from human development occurring in multiple biological and social and cultural contexts.

The life course perspective itself arose from within the historical context of human development research. In that context, it is understandable that life span development was not one of the four original factors identified by Elder in earlier work. It was later posited as a fifth, underlying principle (Giele & Elder, 1998; Tølbøll, 2015) as accumulating health research evidence showed effective preventive health measures in later years required knowledge available only from life-course studies of early influences and behavior patterns (Schulz & Heckhausen, 1996).

Ongoing life course research findings indicated human development and aging were life-long processes. It became clear that late-life adaptation patterns often linked back to formative development. As examples: adult women increasingly benefit from social integration and multiple role activity in later years (Moen, Dempster-McClain, & Williams, 1992); young adults commonly and substantially change work orientation during early years in the labour force (Johnson, 2001); and biomarkers of pre-disease pathways extend back to the formative years (Singer & Ryff, 2001). These developmental records were found to be essential for explaining such behavioral continuity and change over the life span (Elder, 1999). Taking a long-term perspective allowed researchers to reveal this developmental process, the connection and interplay between social change and individual development (Giele & Elder, 1998; Elder, Johnson, & Crosnoe, 2003; Elder & Shanahan, 2006; Alwin, 2012; Tølbøll, 2015).

Key components of the life course

Roles and role configurations, trajectories and transitions, and pathways are three conceptual components of the life course perspective. Each is summarized below.

Roles and role configurations

Roles are basic descriptors of social life; they refer to the “social expectations persons in given social positions have regarding their own behavior and the behavior of others” (Macmillan & Copher, 2005, p. 859). Individuals enact discrete social roles associated with routine behaviors, behavioural expectations, and defined resources within social institutions like marriage, family, school, or work (Macmillan & Copher, 2005). Role configurations refers to age-specific “matrices of discrete social roles that individuals occupy at given points in the life course” (Macmillan & Copher, 2005, p. 859; Macmillan & Eliason, 2003). It is important to note that role configurations – e.g., working parent – requires contextualizing multiple social roles while recognizing the unique meaning of any one role is dependent upon the presence or absence of other roles (Macmillan & Copher, 2005).

Role trajectories and transitions

Life course dynamics are made up of interrelated concepts of role trajectories and transitions (Elder, 1985; George, 2009). Trajectories are the temporal continuity of roles or experience that vary in duration (Macmillan & Copher, 2005). They are situated within historical, cultural, and institutional contexts, experienced as a succession of roles and experiences occurring in individual lives, and perceived as “... long-term patterns of change and stability” (George, 2003a, p. 162). The life course perspective is comprised of four key trajectories: family, learning, employment, and community. Events and transitions along each trajectory change an individual’s role responsibilities at particular points and in particular trajectories (Elder, 1994). Transitions mark the beginning and at the end of trajectories. Typically short in duration, they are indicators of a change in state – like moving to a new role or ceasing an activity (Macmillan & Copher, 2005).

Transitions are characterized in terms of timing (when they occurred in the trajectory), their duration, their sequencing within the life course (e.g., early/late), and the density or close proximity of occurrence (George, 1993, 2009; Mortimer, Staff, & Lee, 2005). Role transitions require both social and personal changes in status and identity (Glaser & Strauss, 1971).

Pathways

The life course is not just comprised of multiple trajectories and transitions. Rather these interconnections or intersections aggregate to indicate general structures of the life course, pathways over the life course (Macmillan & Eliason, 2003). According to Elder (1985; p. 32-33):

“Life course dynamics arise in part from the interplay of trajectories and transitions, an interdependence played out over time and in relation to others. Interdependence emerges from the socially differentiated life course of individuals, its multiple trajectories, and their synchronization. The interdependence ... is also expressed in the concurrence and overlap of transitions along different pathways.”

The idea of pathways conceptually integrates role configurations with transitions and trajectories. Pathways reflect patterns in the order and timing of multiple interconnected social roles over time; they identify particular routes in/out of social institutions (Macmillan & Copher, 2005). At the same time, pathways capture the dynamic unfolding of multiple role trajectories and the interconnections between multiple role transitions over time (Macmillan & Eliason, 2003). In sum, pathways reveal both components and structure of the life course in a given society or social institution. Pathways express the order and timing in the life course by incorporating the dynamic of variable trajectories and meanings of given roles across multiple social institutions and across time (Macmillan & Copher, 2005).

Cumulative advantage/disadvantage

Cumulative advantage/disadvantage (CAD) is an important theoretical approach often used in conjunction with life course research (Dannefer, 2003). Formally, CAD is defined as “the systemic tendency for interindividual divergence in a given characteristic (e.g., money, health, or status) with the passage of time” (Dannefer, 2003, S327). A key factor in cumulative advantage/disadvantage research is heterogeneity that emerges in later life as a result of childhood exposure to risk and adversity (Dannefer, 2003; O’Rand, 2002, 2006; Diprete & Eirich, 2006). Early events and outcomes are magnified with age, resulting in “the unequal provision of physical, social, and economic resources by parents to their children conditions lifelong patterns of inequality” (O’Rand, 2006, p. 149). Initial economic advantage or disadvantage during one’s upbringing is a robust predictor of health trajectories (O’Rand, 2002). The life course perspective provides key conceptual elements for analyzing cohort processes, age-related social-structural processes, and institutional processes together. With it, researchers have applied a cumulative advantage/disadvantage lens to questions of individual and population health (Dannefer, 2003).

Life course applications in health research

A life course perspective is particularly relevant to health outcomes research (George, 1993). Developmental principles of the life course suggest health status is impacted by multiple, long-term trajectories encompassing multiple roles and extending from childhood to late life. Health inequalities may be generated over time by exposure to multiple sources of adversity and disparate access to resources and opportunities (Ferraro & Shippee, 2009; O’Rand, 2002, 2006). The life course perspective provides a theoretical lens to integrate macro-, meso-, and micro-level factors that interact to shape individual life course and health outcomes (George, 1993; Dannefer, 2003). For example, health policy and funding (macro level; historical context) can impact the unequal allocation of health care resources in a community hospital (meso level) which in turn can influence

the agentic and biographical elements of individuals' health activities and outcomes (micro level).

The stress process

Stress and stressors permeate all areas of life. Hans Selye (1956) was among the first researchers to look at the physiological effects of noxious stressors in laboratory animals. By 1967, psychosocial research on stress actively began with the publication of Holmes and Rahe's weighted checklist of major life changes. Since then, causes, attenuations, and effects of stress are well-studied across many research domains. From a sociological perspective "stress or stressor refers to any environmental, social, or internal demand which requires the individual to readjust his/her behaviour patterns" (Thoits, 1995, p. 54).

Pearlin et al. (1981) first described the 'stress process model' and it has become the predominant sociological model to explain the effects of stress on mental and physical health. With revision over time, it has become the distinctive sociological model of stress capable of embracing the complexity of life (Avison, Aneshensel, Schieman, & Wheaton, 2010). The stress process model indicated individuals of different social statuses were exposed to different levels of stress and that resulted in differential physical and mental health related outcomes. Conceptually, the stress process model identifies three main components - sources of stress, mediating or moderating resources, and manifestations or outcomes of stress. There were two key sources of stress – life events (causing stress as they occur at a single point in time) and life strains (manifestation of stress that develops over a longer period of time). The social structural determinants of psychosocial resources like mastery, coping, and social support, were protective factors that mediated or moderated the relationship between stress and health outcomes. Other important social and economic statuses also influenced outcomes. Pearlin et al. (1981) asserted "there are several junctures at which the mediators can conceivably intervene: prior to an event, between an event and the life strains that it stimulates, between the strain and the diminishment of the self-concept, or prior to the stress outcome." (p. 341)

The effects of stress can be direct or indirect; it is equally important to consider both possible outcomes (Pearlin, 1989). Manifestations of stress may be physical and/or mental, with local or global effects (Davie, 2014). In 1999, Pearlin elaborated three underlying assumptions in the stress process model. First, the stress process is dynamic; changes in one component of the model produces change in other components. Second, social stress is a typical manifestation of ordinary life. Stress arises from everyday social roles and is experienced in typical social contexts. Third, stress originates in the social world. This emphasis on the social context of life differentiated the sociological study of stress from historical or biological foundations of the stress paradigm.

Mediators and moderators in the stress process

Mediators and moderators are key components of the stress process model. They help us understand and explain the differential vulnerability of individuals to stressful events (Pearlin, 1989).

A mediator is an explanatory variable that identifies and explains a relationship between two other variables. The mediator governs the relationship between the two variables and explains the nature of relationship between those variables. Often, investigating the mediating effects of psychological resources in the stress process allows researchers to explain vulnerability of individuals affected by stressful events (Pearlin, 1989).

A moderator is a variable that can increase or decrease the strength of, or change the direction of, the relationship between two other variables. The relationship in question pre-exists any moderator/mediator effect. Moderating effects of the same psychological resources buffer individual responses to the stressful event (Turner, Taylor, & Van Gundy, 2004).

Mediating effects of psychological resources have received considerably greater research focus than moderating effects (Matthews, Gallo, & Taylor, 2010; Schnittker, 2007). Turner, Taylor, & Van Gundy (2004) identified only a very small body of literature that supported a buffering or moderating hypothesis for the stress process. The difference

between ‘mastery’ as a mediator or moderator of response to stress may lie in the contextual application of the construct in the study.

Mediators and moderators are comprised of diverse psychological resources including concepts like social support, coping, mastery, self-esteem, self-efficacy, purposefulness, resilience, and autonomy (Aneshensel, 2009; Avison & Thomas, 2010; Kiviruusu, Huurre, Haukkala, & Aro, 2013; Pearlin, 1989; Pearlin et al., 1981, Pearlin, Schieman, Fazio, & Meersman, 2005; Romero-Moreno et al., 2011). These psychological resources can impact at any point in the stress process, but commonly intervene between the onset of a stressor and the manifestation of stress in that situation (Pearlin et al., 1981).

Attenuation effect depends on individual agency and access to important psychological resources (Turner, Taylor, & Van Gundy, 2004).

Cumulative stress

Traditional stress process research differentiated between stressful life events and chronic sources of stress (George & Lynch, 2003; Turner, Wheaton, & Lloyd, 1995). A stressful life event represented an undesirable event that occurred at a specific point in time (e.g., death of a loved one). Chronic stress referred to ongoing exposure to sources of stress (e.g., ongoing financial stress). Chronic exposure to stressful life events often leads to a buildup of stress, or stress proliferation (Pearlin, Aneshensel, & LeBlanc, 1997). Current stress process research jointly considers the impact of life events and chronic stressors. The overall stress burden for an individual is the accumulation of chronic stressors and life events (Turner et al., 1995).

Psychological coping

Psychological coping plays a central role in the stress process model, mediating or moderating the impact of stressors. Coping fundamentally refers to “the things that people do to avoid being harmed by life-strain” (Pearlin & Schooler, 1978, p. 2) and has been conceptualized two ways in the literature. First, coping has been presented as a static personality trait that is relatively constant over time or as a contextual process that

differs depending on the specific stressor (Lazarus, 1993; Lazarus & Folkman, 1980; Somerfield & McCrae, 2000; Penacoba-Puente, Carmona-Monge, Marin-Morales, & Naber, 2013). Second, Lazarus and Folkman (1984) argued coping is a constantly changing process comprised of cognitive and behavioral aspects. Coping consists of thought and action taken by an individual in response to a specific stressful event. Coping is in essence protective, guarding the individual from psychological or physical harm by an outside stressor (Monat & Lazarus, 1991).

Three decades of coping research differentiated two dominant presentations: problem-oriented coping occurs when the individual acts to directly alter the stressor or distance themselves from the event; and emotion-oriented coping occurs when individuals alter their emotional reaction to the stressor without removing the condition (Folkman & Lazarus, 1980; Billings & Moos, 1984; Folkman, Lazarus, Dunkel-Schetter, DeLongis, & Gruen, 1986; Monat & Lazarus, 1991; Roybyrne et al., 1992). A third sub-category of coping has emerged – avoidance (denial), occurring when individuals cope by way of distraction or social diversion (Kohn, Hay, & Legere, 1994; Roesch et al., 2005; Taylor and Stanton, 2007). All coping mechanisms may have positive and negative influences on the individual (Monat & Lazarus, 1991).

Social support

Supportive relationships play an important role in health outcomes. Lazarus and Folkman (1984) identified that “people will have better adaptational outcomes if they receive or believe that they will receive social support when it is needed” (p. 259). Social supports have been shown to buffer the effects of stress, protecting individuals from negative influences of stressors (House, 1981; Ryan, 2001). Andrews, Tennant, Hewson & Schonel (1978) found psychological impairment in a crisis was associated with low levels of expected support from friends, family, and neighbors. Pearlin et al. (1981) illustrated social support and coping buffered depression, low self-esteem, and loss of mastery after job loss. Bass, Noelker, and Rechlin (1996) found several types of service support (i.e. personal care and household assistance) significantly buffered caregivers from

psychological and social stress. Mediation may reduce or prevent stressful appraisal of the situation and increase the ability for appropriate situational responses (Maguire, 1991; Monat & Lazarus, 1991; Sarason, B., Sarason, I., & Pierce, 1990).

Social supports have been differentiated using two modes and three functions (Zarit, Pearlin, & Schaie, 1993). Modes are informal and formal sources of support. Informal sources of support include spouse or partner, family, friends, neighbors, or other non-professional resources. Formal sources include professionals like social workers and related professional organizations. Functions are emotional, informational, or instrumental in nature (Gottlieb, 1981; Lin, 1986). It is important to note these functions are not mutually exclusive (Lazarus & Folkman, 1980). Emotional (expressive or esteem) support provides a sense of acceptance, belonging, and personal value to enhance the ability to cope with stressors. Informational (advice or appraisal) support helps define and understand the problem to improve coping abilities. Instrumental (tangible) supports like financial assistance and material resources provide respite and additional time to address the stressful event (Ryan, 2001).

The considerable body of research on the buffering effect for coping and social support showed mixed results (Baron & Kenny, 1986; Cohen & Wills, 1985; Thoits, 1982, 1986). Much of the confusion stems from theoretical and methodological inconsistencies across studies (George, 2003b). Social support and coping concepts may be specified in the same manner and are sometimes applied interchangeably, and researchers have diverse ways of defining and measuring the social support variable (Cohen & Wills, 1985). Another error has been to equate the size of social networks – the number of relationships a person identifies – with social support. What matters is that individuals perceive and receive social support from important others in their network. A large social network does not compensate for lack of support from others in that network (Lazarus & Folkman, 1980).

The stress process in family caregiving research

Sociological research has shown parents of a child with a life-threatening illness like cancer faced a preponderance of chronic strain. Caregiving responsibilities often extended over the longer term. As well, parents worried about their child's future well-being, and emergence of a new or secondary course of illness (Avison et al., 1993). From a stress process perspective (Pearlin et al., 1981), life events (stress at a single point in time) and life strains (stress developing over time) are the key sources of stress.

Contextualizing the caregiving experience of parents when their child undergoes cancer treatment can be challenging. Different cancers, different stages and risks, different personal and system resources all influence short and long term stress for parents and the well-being of their child. The following quote by Sharon Manne, a Pediatric Psychologist, on her liaison work in a Pediatric Oncology Unit captures the lived experience of many affected parents:

The most striking issue to me over the years of doing this work was the immense practical problems and emotional stress placed on a parent of a child being treated for cancer. The parent providing primary care to the child, typically the mother, may reside for months in the hospital, leaving behind her remaining children and her spouse (along with her own sources of emotional and practical support) in a distant city. Hospital staff and other parents become surrogate sources for support for the caregiving parent. Furthermore, caregiving parents are expected to manage their child's complex medical regimens both inside and outside the hospital. For many parents, what is most upsetting is witnessing the suffering of the child they are responsible for protecting. Parents watch their child undergo extremely invasive procedures ... (resulting in) temporary or permanent disfigurement or disability caused by surgery; or lengthy, intense chemotherapy, radiation, or stem-cell transplantation. (Manne, 2005, p. 7375).

In 1997, Pearlin et al. demonstrated the stress process could effectively explain the stress of caregiving. A broader body of literature supports the utility of the stress process in understanding caregiving scenarios (Pearlin et al., 1997; Turner et al., 1998; Aneshensel, Pearlin, & Schuler, 1993, Aneshensel, Pearlin, Zarit, & Whitlatch, 1995; Pearlin, 1992; Skaff and Pearlin, 1992; Skaff, Pearlin, & Mullan, 1992). Collectively, this work introduced the stress process paradigm to health science disciplines concerned with family-based care outcomes (Avison et al., 2010). Two main types of stress were revealed – one emerged with normative child-related tasks (i.e., minor daily hassles) and the other occurred as a result of critical life events like childhood illness or disability. On the whole, findings supported the concept of stress proliferation where one stressor can result in exposure to subsequent stressors in a cumulative fashion. In lived experience, singular life or situational stressors with little individual impact may cumulatively become significant for parents with insufficient social support. Positive engagement in multiple social relationships was important – spouse, friends, and community support moderated the stress of these parents. Key situational or environmental stressors proliferating parental stress include poverty, single parent status, and the presence of multiple younger children in the home (Breslau & Davis, 1986; Crnic & Greenberg, 1990; Kovacs & Feinberg, 1982).

Caregivers of children with a chronic and/or terminal illnesses face a preponderance of chronic strain. Advancing treatment protocols often provide longer life expectancy for many children with life-threatening and debilitating illnesses such as diabetes or cancer. However, the child's health may not normalize even with optimal treatment. These parents face on-going caregiving responsibilities, worries about the child's well-being, future therapies, and the risk of a new, emerging course for the illness (Avison, Turner, Noh, & Speechley, 1993). For children with cancer, parents' perceived level of social support was an "extremely important predictor of distress" (Avison et al., 1993, p. 91). Even though a significant moderating effect was not found, the authors suggested "these resources do seem to play some role in reducing the effects of the chronic strains associated with giving care" (Avison et al., 1993, p. 92).

Merging stress process and life course in research

Historically, the stress process and life course were largely separate research domains (Elder, George, & Shanahan, 1996). Pearlin and Skaff (1996) first suggested principles central to the stress process naturally aligned with life course principles. Together these sociological theories offered a comprehensive model to examine how individuals' exposure to stressors changed or showed differential effects over time. Pearlin et al. (2005) elaborated the key stress process factors that affected health outcomes over the life course. Status attainment was identified as central to better health outcomes over the life course. Health effects of status inequality commonly occur through indirect, differential exposure to stressors and hardships.

From a life course perspective, status attainment is experienced primarily on the education-occupation-economic pathway as a chain of status contingencies, adversities, and benefits (Pearlin et al., 2005). Status-related exposure to stressors may be most conspicuously experienced in the neighborhoods people live. Residents experience a number of ambient stressors that impact multiple roles and spheres of social activity to increase adversity and diminish well-being (Pearlin and Skaff, 1996; Aneshensel and Sucoff, 1996; LeClere, Rogers, and Peters, 1998; Pearlin et al., 2005). Ambient stressors can appear as constrained opportunity structures when great value is placed on achievement. Individuals lagging in status attainment often experience failure and frustration while striving for these socialized aspirations. This stress is internalized as negative self-evaluations that impact self-image and reduce agency (McLeod & Nonnemaker, 1999).

Chronic hardship, role strain, and discrimination are also rooted in status location and recognized by continuity or repetitiveness across the life course (Pearlin et al., 2005). Consequences of chronic or recurring stressors may be particularly severe when they surface within major social domains like family, income, or employment (Avison & Turner, 1988; Pearlin, 1983; Wheaton, 1999). Stressors involving ascribed statuses like race and gender challenge important social rights and threaten role identities. Continuous

and repeated stressors are most likely to cumulatively increase the allostatic load – the biological burden placed on the organism when responding to hardships (Seeman, McEwen, Rowe, & Singer, 2001).

The concept of stress proliferation emerged from the observation that serious event stressors or chronic role strains tended to produce additional stressors (Pearlin, 1989; Pearlin et al., 1997). Pearlin et al., (2005) identified three situations where stress proliferation has long-term consequences for health: early trauma, the timing and sequencing of transitions, and the disruption of roles and statuses.

Early traumas are potent events in stress proliferation research. Traumas are distinguished from other stressful events by their magnitude of impact (Wheaton, 1994) and an often sudden, critical onset (Norris, 1990). Stressors may stem directly from the trauma or emerge later as secondary stressors that exert their own harmful effects on individual health and well-being (Kessler & Magee, 1993). Proliferated stressors may emerge as additional trauma, chronic strain in important life domains, or other stressful life events. Early traumas and their proliferated stressors are disproportionately more for individuals with education and financial disadvantages, explaining some of the health disparity related to socioeconomic inequality (Pearlin et al., 2005; Turner & Avison, 2003; Turner et al., 1995; Wheaton, Roszell, & Hall, 1997).

Timing and sequencing of early transitions play a key role in long term well-being across many life domains. Transitions that were undesired and involuntary often negatively impact personal well-being (Thoits, 1983; Pearlin & Mullan, 1992). Critical early life transitions most often lead to chains of adversity that are difficult to alleviate. Deviation from normative timing and sequencing of pivotal roles and statuses – like transition out of formal education and into the labor force – restricts access to vital opportunity structures, effectively preventing a smooth and timely transition to other roles and statuses such as well-paid employment in a chosen career. Disorderly transitions tend to push individuals to lower socioeconomic strata and heighten the risk future stress proliferation in multiple life domains or pathways. Collectively, proliferated stress

imposes a large allostatic load¹⁰ on the organism resulting in later-life manifestations of stress-related diseases (Pearlin et al., 2005; McEwen and Seeman, 1999).

Life course disruptions involving established roles, statuses, relationships, and activities are themselves stressors but may also proliferate to social domains beyond the origin. Such disruptions are typically undesired, unanticipated, and involve loss. Family caregiving is a closely studied situation where secondary stressors are known to emerge over time and impact other life domains or roles (Pearlin, Mullan, Semple, & Skaff, 1990; Aneshensel et al., 1995; Pearlin et al., 1997). Caregiving is an emergent role, neither fully anticipated or prepared for. However, some secondary consequences of caregiving will more likely surface in the lives of persons with less advantaged statuses – financial strain for persons with lower incomes or individuals struggling to balance demands of outside employment with those of caregiving. Not only the principal caregiver is exposed to the stressors associated with the role, the stressors can penetrate the lives and roles of others nearby (Pearlin, Pioli, & McLaughlin, 2001; Semple, 1992). Here stress proliferation is configured differently, not only spreading across time for the caregiver, but also spreading out to others sharing a role set (Pearlin et al., 2005).

From a life course perspective on stress, individuals move through the life course on unique pathways of change. These trajectories of change are grounded in childhood experiences, plus the person's current position in the life course and exposure to recent and ongoing stress. Lives are restructured across time by social context and the occurrence, timing, and sequence of stressful events and situations (Gotlib & Wheaton, 1997; Umberson, Williams, Powers, Liu, & Needham, 2005).

¹⁰ Allostatic load is the accumulated physiological consequences (i.e., wear and tear on an individual's body) with exposure to repeated or chronic stress (McEwen, 2003).

Chapter 2

Methodology

This chapter presents the qualitative research methodology used to examine the nature of fathers' experiences as they parent a child with Retinoblastoma. Mothers – spouses and partners where possible – were considered the control group and interviewed using an identical interview instrument to provide comparative data for analysis. There is a small body of previous material on fathers in the psychosocial oncology research literature. Most information was, and continues to be, derived from survey approaches. Little comparative qualitative research has been completed; of the literature, a dearth considered sociological perspectives like the stress process to examine how parents and couples coped when their young child was diagnosed with cancer. As well, explorations of individual, family, and marital dynamics with infant and childhood cancers continues to be weighted towards mothers as participants. This inherently creates a knowledge gap around fathers' experiences, minimizing a sociological understanding of stress as it relates to their experiences, challenges, and necessary supports required for optimal coping during the highly challenging period of diagnosis and active treatment for their young child's cancer. Following sections in this chapter contextualize the study; summarize the research design, research questions, and method of choice; identify the target population, describe sampling and recruitment activities; review the interview process along with handling, preparing, and analyzing the data; address data quality issues; and identify research ethics approval.

Reflexivity statement

Reflexivity is optimally a continuing process of reflecting on and tracking researcher assumptions and decisions that could be influencing a research project. It can be thought of as a self-portrait of potential experiences and bias, identifying personal factors that might influence decisions and interpretations in the research process. Issues like who to sample out of a target group, the questions asked of participants, or even selection of

respondent quotes for thematic presentation. This approach is critical for hard-to-reach populations and where the researcher has a close engagement with both the subjects and the data. There is the possibility that a personal stance could impact interpretation in these situations (England 1994; Bourke 2014; Turner 2016).

The development of this research project emerged from formal participation in a three-year collaborative, interdisciplinary research program arranged between my supervisor in the Department of Sociology at the Western University in London and the Head of the Retinoblastoma Program at the Hospital for Sick Children in Toronto. As a Sociology doctoral student, I was immersed in a specialized clinical research and treatment team focused on Retinoblastoma, a genetic eye cancer. While I had prior clinical health research experience, this was a unique graduate placement arrangement for both organizations and an entirely new experience for me. A sociology graduate student embedded in this basic science and clinical medicine team was a first too. It was also a unique, shared experience for a large medical team comprised of clinicians, residents, International fellows, graduate students from a variety of medical disciplines, other clinical researchers and specialists, and software developers. Integration included expectation that I regularly attend related medical education sessions with fellows and observe at weekly clinical case review meetings. An open office configuration enhanced opportunity for interaction, discussion, and learning.

My placement did not begin with qualitative research or even sociology. Prior work experience meant my first project focused on optimizing the user interface for a new clinical cancer database (eCANCER), developed by the Health Informatics Research (HIR) Department at Princess Margaret Hospital. The Head of Retinoblastoma at the Hospital for Sick Children (HSC) was also the HIR Director at Princess Margaret Hospital. As a result of that linkage, Retinoblastoma was the pilot cancer for the eCANCER platform development. In retrospect, that initial decision – using Rb for the pilot – was the first phase of an interdisciplinary experiential learning experience from which this sociological research project emerged some 3 years later.

The user interface project introduced me to the science and clinical thought around this rare and complex disease – diagnostic classification scheme, current treatment protocols, emerging research, genetic factors and related diagnostics. During this period, I collaborated on quantitative survey research that suggested a positive relationship between visual aids (a clinically-relevant visual timeline) and greater parent understanding of complex treatment risk and outcome (Panton et al., 2009). This research was completed with parents having a child in active Rb treatment. From that work, I believed good visualizations could support and strengthen parents' timely decision-making and fully informed consent around treatment.

My position on the overall effectiveness of this communication tool for parents changed as eCANCER^{RB} was piloted in Ophthalmology clinical settings at HSC, including the surgical waiting room and dedicated Rb operating room. This venue was a naturalistic setting for six months of weekly direct observation of parents dealing with acute and complex medical information to make treatment decisions. I observed the real-time clinical application of the eCANCER^{RB} to visually summarize and communicate case information to parents which did not seem to have the same impact as the Panton et al., (2009) study suggested. Parents politely received the information but very much preferred one-on-one time with the attending surgeon to really understand their child's status. So, was there was something about that personalized interaction with clinicians that helped parents understand and cope, something more than just knowledge translation? For me, larger research questions were forming – was the observed gap (research versus my clinical observation) an error or a result of my position as an outsider to the inner workings of acute-care clinical practice? What could I be missing or what implicit bias might I be imposing on the situation? Was bias present in the quantitative survey research design, in effect providing findings clinicians implicitly believed were true? Could well-designed qualitative research address the gap between my observations and prior quantitative findings? My ongoing discussion and interaction with clinical team members indicated a general recognition of the one-on-one advantage and provided what they could with limited time resources. Database tools like eCANCER^{RB} were viewed as

having potential to enhance communication of complex information to parents, perhaps more effectively than through face-to-face meetings with clinicians. The Retinoblastoma Team worked within a family-centred model of care and were heavily reliant on Nurse and Social Work professions to cover much of the extended information and emotional support to parents. So, was parent understanding of this complexity actually rooted in, or impacted by, comprehensive psychosocial support and not just effective knowledge translation? How large was that impact and what were outcomes for parents who did not access these services? How could parent engagement be optimized? Participating in annual fundraising events and family learning days provided more direct contact with parents who held a strong sense of community as a result of these non-clinical experiences. How important might these activities be to increasing parent and survivor engagement over the longer term?

Further, the quantitative research described above simply replicated the existing sample bias towards mothers in the psychosocial oncology literature. What about fathers? Available accounts of their experiences seemed superficial and insufficient except for practice knowledge shared by the RB Team Social Worker. Was it as simple as fathers attending appointments with their partner but did not participate in research? My observations suggested there were far more mothers than fathers in the waiting room on a regular basis. Were there differences between fathers that attended and those that did not? How would I go about defining father categories for this kind of research? Ongoing interaction with the clinical team in various hospital settings confirmed some of my observations here but offered no immediate research solution. I relied on broad consumption of sociological gender literature for insights and guidance.

An invitation to join the National Retinoblastoma Strategy (NRbS) intersected with my database and clinical observational research at HSC. Pan-Canadian group membership included expertise in the fields of ophthalmology, oncology, genetics, social work, nursing, and advocacy. Rb survivors and their families were also active members. Three years of meetings, consultations, and personal communications with stakeholders

extensively shaped and solidified my sociological thinking and planning for this dissertation research. As lead author for the Psychosocial Care and Access to Services section (National Retinoblastoma Society, 2009), I collaborated with Rb survivors and parents, a policy expert, a Social Worker with qualitative Rb family research experience, and a Clinical Psychology doctoral student. Broadening my focus beyond the hospital clinical setting by interacting with psychosocial-focused professionals provided strong and consistent support for a qualitative research approach. I wanted this research to impact clinical psychosocial practice; consensus from this group pointed towards a strong need for insights to improve coping and social support strategies for Rb parents, in both active care and follow-up. Rb parents certainly experienced multiple stressors and the stress process seemed a good model for individuals. Anecdotal clinician reports of marital stress and breakdown early post-diagnosis also indicated a dyadic stress model was worth considering. The stories of adult Rb survivors strongly suggested a researchable life course perspective if I could find a way to connect with those individuals as they were no longer connected to the hospital setting. Support for father-focused research was encouraged by the broader group, including clinicians from other roles and treatment centres. One Ophthalmologist had completed exploratory focus groups with fathers and found feedback helpful for clinical practice. By the time the National Retinoblastoma Strategy wrapped up, the HSC team was fully supportive of this research project. A comprehensive sociological literature review literature was completed, five research hypotheses were defined, and the interview instrument was under completed. Literature review for possible strategies for categorizing fathers was ongoing. Reflexive practice continued throughout the interview process, including regular supervision and writing field notes from in-home and hospital interviews. This extended throughout the iterative coding process to ensure consistency across categories and themes.

Research design

Sociological research of this nature can be classified as a natural experiment. This is observational research, occurring in natural settings and taking advantage of a naturally

occurring event outside the control of the researcher (Dunning, 2008, 2012; Dawson & Sim, 2015). In natural experiments, individuals and groups may be differentially impacted by the event but researchers do not have the ability to randomly assign to groups. In the case of Retinoblastoma, exposure closely resembles random assignment as incidence is governed by random genetic mutation in the proband (first individual in the family history to have the disease). With history, the likelihood of disease can be much greater so assignment is less than random, but still beyond experimental control. It would be impractical and unethical to attempt experimental research designs of this nature. Observational research in natural settings can have high utility in social research when a clearly-defined event impacts a specific and well-defined subpopulation in a manner that changes can be plausibly attributed to the event. As a result of a natural event, individuals or groups may be differentially exposed to conditions that can be exploited to answer particular research questions (Gylmour, 2006; Messer, 2008; Dunning, 2008, 2012; Dawson & Sim, 2015).

Interpretive description as method of choice

Over the last three decades, qualitative clinical health research designs have increasingly been implemented to provide contextual insights and answers to compelling and complex questions. Researchers traditionally used a broad range of epistemological perspectives such as phenomenology, ethnography, or grounded theory (Thorne, Kirkham, & O'Flynn-Magee, 2004). Researchers asking applied health and clinical research questions were often constrained to fit their research to the objects and methodological rules of disciplines as diverse as sociology, anthropology, or philosophy (Morse & Chung, 2003). This practice gap resulted in a move away from orthodox qualitative methodological practices and was most evident in qualitative nursing research (Morse, 1994). Emerging practices articulated distinct approaches to research design, approaches that fit complex experiential questions asked by these applied health researchers.

Thorne, Reimer Kirkham, & MacDonald-Emes (1997) developed interpretive description as one approach to investigate clinical phenomenon. According to Thorne et al. (2004),

“[t]he foundation of interpretive description is the smaller scale qualitative investigation of a clinical phenomena of interest to the discipline for the purpose of capturing themes and patterns within subjective perceptions and generating an interpretive description capable of informing clinical understanding.” (p. 3)

Studies of this nature commonly engage small samples and collect data through multiple sources such as interviews, participant observation, and documentary analysis. This minimizes what Sandelowski (2002) labels as a naive overemphasis on interview data at the expense of comprehensive and contextualized interpretations of the clinical question.

Interpretive description has been described as a non-categorical method of research and critiqued for risk of ‘method slurring’, the blurring of distinctions between established qualitative approaches (Baker, Wuest, & Stern, 1992; Morse, 1989). Some researchers have also identified the lack of epistemological and methodological grounding as a weakness in this generic qualitative method (Caelli, Ray, & Mill, 2003). However, Thorne et al. (1997, 2004) argue a strong philosophical alignment with interpretive naturalistic orientations, approaches embracing the constructed and contextual nature of human experience while acknowledging and allowing shared realities. Three axioms of naturalistic inquiry delineated by Lincoln and Guba (1985) underpin interpretive description:

- a. Social reality is complex, constructed and contextual, so ultimately subjective. The result is multiple, co-existing realities that must be studied holistically.
- b. The inquirer and the object of inquiry interact and influence each other; the knower and the known are inseparable.
- c. No a priori theory can encompass multiple realities encountered in dynamic, lived settings. Theory must emerge or be grounded in the data.

The authors argue that, while the techniques for data collection and analysis may vary within the interpretive description method, the coherent epistemological foundation of

naturalistic inquiry are embedded in this approach and distinguish it from assumptions characteristic of method slurring.

Interpretive description stands apart from some traditional qualitative descriptive approaches in that it assumes researchers go beyond description, exploring meanings and explanations that may be applied to clinical practice. Inductive analytic approaches allow researchers to understand clinical phenomena in a theoretically useful manner. Simply stated, “interpretive description provides direction in the creation of an interpretive account that is generated on the basis of informed questioning, using techniques of reflective, critical examination, and which will ultimately guide and inform disciplinary thought in some manner” (Thorne et al., 2004, p. 4). Research design strategies borrow strongly from ethnography, grounded theory, and naturalistic inquiry while incorporating phenomenological approaches to data collection. Sampling is purposive and often theoretical, indicating the researcher has a priori awareness or at least the expectation of emerging variations in the target phenomenon.

The product of interpretive description is two-fold: the coherent conceptualization and description of themes and commonalities and an accounting of individual variation across subjects. Generating new theory or original explanation is beyond the scope of interpretive description (Sandelowski & Barroso, 2003; Thorne et al., 2004). However, capturing particularly strong metaphors and images offers researchers an avenue to enhance articulation –

“... about what is common within a clinical phenomenon. In the disseminated research report, such a claim would be rendered accessible to the practice of the discipline for the purpose of informing clinical reasoning, extending the available insight for practice decisions, and creating a sense-making structure for the eccentricities and variations that inevitably occur in the real world of health care application.” (Thorne et al., 2004, p. 4).

The interpretive aspect takes on a critical perspective, providing health care disciplines with "... a backdrop for assessment, planning, and interventional strategies" (Thorne et al., 2004, p. 4) that follow practice standards of evidence, logic and ethics. Sandelowski & Barroso (2002) indicated good interpretive description makes sense of something clinicians ought to understand. Themes and metaphors provide mental heuristics linking new knowledge to enhance clinical practice as if gains had been acquired through both clinical and reflective practice. In sum, analytic processes of interpretive description provide the utility and quality of information generated to enhance clinical practice knowledge and care outcomes (Thorne et al., 1997; Thorne et al., 2004; White & Multiple Sclerosis Society of Quebec, 2002).

Acquiring research ethics approval

This study received ethical approval from Research Ethics Boards at both the Hospital for Sick Children (HSC) in Toronto, Ontario and Western University in London, Ontario.

The initial application was made to the Research Ethics Board for The Hospital for Sick Children in Toronto, where this researcher was completing a graduate research placement on the Retinoblastoma Team in the Department of Ophthalmology. This research was based out of HSC in Toronto. A signed copy of the final Approval & Terms of Agreement REB#1000012956 from the Hospital for Sick Children was provided to the Research Ethics Board at Western University to comply with the institution's protocol for approving external research having already received REB approval at another recognized institution.

This study underwent internal peer review from clinicians in the Departments of Ophthalmology and Social Work at HSC as part of the REB application, review, and revision process. The risk classification for this project was 1B – a prospective

observational study requiring adverse events reporting and annual REB reports.¹¹ The Principle Investigator (PI) for this study was the Head of Retinoblastoma at The Hospital for Sick Children and this researcher's direct supervisor for all research projects and associated learning in hospital and clinical settings. This researcher was co-investigator on the REB application. The PI assignment to a senior supervisory clinician was regular practice for all student research conducted in this active care clinical setting. This researcher participated in multiple, sometimes concurrent REB-approved 1A retrospective or clinical file review projects using Rb patient records before and during the development and data collection phases of this project. Similarly, Rb parent survey research projects (assigned to medical Clerkship students with methods support from this researcher) had information derived from clinical record reviews. All these projects involved active use of Rb clinical records, supervised by the same Principle Investigator, and overlapped with this research endeavor. As described in Chapter 2 'Reflexivity statement', this researcher had regular and ongoing exposure to patient records through participating in a variety of formal research projects and informal community activities. The end result was active and passive exposure to a variety of case-related and contextual information that could not be excluded from this project – I could not forget clinical information I already knew just for this project. Regular supervision with the PI guided the ethical use of this information to prepare for interviews and triangulate for quality checks.

Approval of this research addressed structure and content of the invitation letter; confidentiality of all information shared; secure storage and destruction of data; and the risks and benefits of participation. Obtaining written informed consent was required prior to each in-person and telephone interview and a separate written consent was required to audio-record each session. Participants were provided access to the Rb team Social

¹¹ Research Ethics Board for The Hospital for Sick Children Approval & Terms of Agreement REB#1000012956, p. 10, Dr. Brenda Gallie & R. Downie.

Worker for support should participation in the interview cause them any distress. Parents were also provided contact information for the Principle Investigator.

Developing the interview instrument

At a design level, three types of qualitative interviews are possible: structured, semi-structured, and unstructured (Fontana & Frey, 2005). The primary data collection strategy for this study was in-depth, semi-structured interviews. Each participant had opportunity to provide narrative responses to research questions in open conversation, sharing experiences to their individual level of comfort. Together, the interview instrument and process kept the content focused on specific research questions and maximized data gathering across all participants. Careful pre-planning of the set of questions and probes supported greater completeness and dependability of participants' responses (Patton, 2002). The semi-structured format also helped control inherent bias of the single-researcher approach, thereby supporting quality research as measured by trustworthiness and rigour of the data.

Conceptualization of content and questions for the research project and interview instrument followed an iterative process that began after the start of a formal research placement on the Retinoblastoma Team at the Hospital for Sick Children in Toronto. Participating in early eCANCER^{RB} database development provided the researcher with a conceptual understanding of core clinical, diagnostic, and treatment aspects of Retinoblastoma. Providing research support to clinical research projects with researcher-clinicians and Rb fellows provided a whole new level insight into parental experience of their child's clinical treatment through direct contact with parents of children undergoing active treatment at the hospital. This first-year experience coincided with reading for comprehensive exams in sociology of health and gender and overlapped with membership on the Canadian Retinoblastoma Strategy group. The result was a proposal to complete qualitative sociological research focused on fathers of children treated for Retinoblastoma. Theoretically, the research was grounded in a masculinities perspective (e.g., Connell, 1995; Kimmel, Hearn, & Connell, 2004) and explored both fathers' role

strain and identity (e.g., Cooper, 2000; Robb, 2004) and the stress impact on the marital dyad (e.g. Brody & Simmons, 2007) as a result their child's Rb diagnosis and treatment.

Content and structure of the first draft was based on the researcher's understanding Rb disease and treatment processes, sociological stress and coping models, a comprehensive review of the psychosocial oncology literature, collaboration on the National Retinoblastoma Strategy, ongoing research projects and observation in clinical settings, and personal communications with Rb parents who were stakeholders in these activities.

Operationally, the interview instrument was constructed like a survey instrument with thematic sections and embedded skip logic to bypass follow-up questions that did not apply. Each section had an initial yes/no response screening question – Were you born in Canada? Are you currently employed? As example, respondents who were born in Canada skipped following questions exploring immigration status. Full-time and part-time employment questions were duplicated in the instrument but each respondent answered one set (6 of 12 questions). Respondents not in the labour force at interview time did not receive any of these questions.

Many interview items actually collected contextual or demographic information in short-answer questions like: How long have you held this position? Was this a planned pregnancy? Are you (CHILD)'s biological mother/father? How old were you when (CHILD) was born? As discussed in Chapter 2 'Reflexivity statement', this researcher had supervised and REB-approved access to clinical records and information for research projects and from clinical meetings as a member of the Retinoblastoma Team. Already-known case-specific (medical) information was not asked in the interview and inserted post hoc into the paper form. Parents who were present when their child was diagnosed were not asked the how and when of learning the diagnosis. Similarly, when interviewing the second individual in a dyad common information – How long have you been married? – known from the first interview was not asked of the partner.

The interview instrument contained 81 items or questions. Interviewees actually completed in the range of 40 to 43 questions because of the skip logic. Many of these were short-answer descriptives by design. Depending on the individual and case, 17 to 19 key questions provided the bulk of interview narrative for this research project. After the consent process, interviews with mothers averaged 70 to 90 minutes. Father interviews were shorter, averaging just under one hour.

The initial questionnaire draft was reviewed by this researcher's Western supervisor; recommended changes were completed. The second review was completed by two Fellows on the Retinoblastoma Team; recommended changes were made to the instrument. Revision three incorporated feedback provided by the Head of the Retinoblastoma Team. Revision four was based on feedback from the Rb Team Social Worker and a final review by the researcher's Western supervisor (see Appendix C).

As a final quality check, the researcher pre-tested the questionnaire by completing four interviews with parent volunteers who were key stakeholders for the National Retinoblastoma Strategy. These individuals held comprehensive lay knowledge of Retinoblastoma and were asked to provide general and specific feedback on the instrument – active feedback on clarity, brevity, and purposefulness of the instrument; comments on item construction and relevance; the interview process, the flow and length; and to indicate any moments or topics that elicited particularly stressful feelings. This member check also confirmed that this researcher satisfactorily captured what respondents said and meant in interview statements. This evaluative feedback resulted in no substantive changes to the instrument. The four interviews were excluded from the final analysis. The comprehensive review process provided an ethical and sociologically sound instrument that was contextually sound from clinical and medical perspectives.

The final (fourth) revision was accepted as submitted with the Research Ethics Board application.

Recruiting participants for this study

Outside of a mandatory disease-specific registry there is no way to accurately document the true Canadian Rb population. There is no such Rb registry, so no comprehensive single-source data set was available for study recruitment. The initial research plan was to use HSC Ophthalmology clinical records to recruit for the study. While this was a robust sample, it did not address essential research design elements of national representativeness and a life course perspective experienced by parents of older adult survivors. Ophthalmology active clinical care and follow-up records were insufficient for the intent of this qualitative study which was to interview a representative national sample of Rb parents whose child was at any stage of treatment, follow-up, or adult after care.

An important consideration for this study was that national delivery of Rb care is segmented by disease presentation and case complexity. This dynamic meant there was no single source or representative set of patient records to use for study recruitment. Bilateral and complex unilateral cases were all treated at HSC. Simple unilateral patients may receive treatment only in the secondary care centres closest to their home, never showing in HSC records. Some tertiary-care patients moved to secondary treatment centres (closer to home) as their case stabilized. HSC clinical records were no longer current unless there was further consultation between clinicians at the centres. At 18 years of age, all survivors (and families) moved to the adult care system and disconnected from pediatric health care services. Past attempts at multi-site recruiting have shown to be resource-intensive and resulted in samples skewed to active treatment populations. Older Rb families and those living farther from treatment centres had very low participation rates. Accessing clinical patient information from other institutions was also challenging, even in externally-funded projects.

It was important to consider time and disease presentation factors when designing a recruitment strategy to extend understanding of Rb parent stress experiences beyond the acute care setting. The psychosocial literature has largely considered acute stress impacts of diagnosis and treatment of childhood cancers (e.g., Gallie et al., 1996) on mothers and families (e.g., Clarke et al., 2003). Long term follow-up has largely been from an epidemiological perspective, documenting population incidence of secondary cancers in Rb survivors (Abramson et al., 1985). There is a dearth of experiential and life course studies in the psychosocial oncology literature. This information gap is most notable for those with complex treatment profiles that may include vision loss as well as secondary cancer risk in later life. Project success meant successfully recruiting parents of older adult survivors for a comprehensive view of stress and coping life experiences.

Purposive sampling (Morse, 1991; Oliver & Jupp, 2006) directed recruitment efforts to participants who were Rb parents of a child diagnosed and treated for Retinoblastoma. Only that experience provided respondents the authority to speak on personal, marital, and family experiences explored in the interview questions. Snowball sampling (Patton, 2002; Oliver & Jupp, 2006), also a type of purposive sampling, emerged as a secondary recruitment in the early strategy to recruit qualified participants. This approach was a result of early study interviewees recommending other affected family members (i.e., affected grandparents) participate in the project. In some cases, individuals received the original email invite from the Canadian Retinoblastoma Society and participated after encouragement from other family members who had completed the interview. Practically speaking, the sample was one of convenience – respondents self-selected to participate in a study interview.

A viable recruitment alternative emerged through this researcher's prior collaboration on the National Retinoblastoma Strategy (NRbS) and participation in annual community events (see Chapter 2 'Reflexivity statement'). The Canadian Retinoblastoma Society (CRbS) offered the researcher an alternative mechanism to recruit participants – namely their email membership list. The Canadian Retinoblastoma Society (CRbS) is a national

advocacy group with membership comprised of Canadian Retinoblastoma families and survivors with lived experiences, from newly-diagnosed to long-term survivorship. The Society sponsored both the NRbS and annual Rb family events. Contextually, CRbS membership was a voluntary partial national ‘registry’ of Rb survivors and families. Self-selection for membership remained a bias with this source but the opportunity to recruit nationally and engage older Rb families for a life course perspective aligned with core study design criteria. The contact list provided a large but non-exhaustive resource of possible participants to recruit. At project implementation, 240 unique member families were available as the target population.

The CRbS President was very supportive of this parent-focused experiential research. With Board endorsement, the President sent the invitation to participate in the research study to all parent/family members by email. The CRbS was a trusted resource to these families, giving the research invitation a higher degree of initial credibility than direct mail recruitment, possibly enhancing the response rate. From prior collaborations, this researcher understood CRbS membership was geographically diverse, offered families with a wide variety of disease/treatment outcomes, and included both older survivors and their families. Therefore, an email recruitment aligned well with core study criteria for participation.

The use of clinical records may have expanded participation for recently-immigrated Rb families living and having a child treated in Toronto. However, earlier clinical research showed families with lower levels of English language proficiency were less likely to engage in any Rb research (Panton et al., 2009). Translation services were not available for this study and email recruitment would offer no advantage to off-set any language barrier in the interview. A multi-language and multi-ethnic research team would seem to be the best avenue to include newcomers in such a research project.

Conducting the interviews

The study was introduced in the body of the email sent by the President of the Canadian Retinoblastoma Society to all registered member families. The attached clear language invitation to participate explained the project, the rationale, and the interview request to parents. Parents could respond by email or telephone to this researcher for more information on the study and arrange to participate in the study if they so chose. Each parent responding by email or telephone and indicating interest in the research was sent the information package by mail or email. This included the invitation letter, the consent to participate in the study interview and the consent to audio-record their session. Mailed packages included an addressed, postage-paid envelop for mail-back of signed consent forms. The researcher appropriately followed-up with a communication to schedule a telephone call with each individual to fully review each consent form and answer all questions. If individuals chose to participate at that point, we selected a time and date suitable for their schedule (sometimes this was months out on their next trip to Toronto and it was usually one parent at a time). Signed consent forms arrived by mail or email (as scanned attachments). Some parents brought signed forms to the interview. Before each interview, I again reviewed consent, answered further questions, countersigned the forms, and provided a signed copy for their record.

The initial research plan was to interview individual parents when they brought their child to the hospital for regular clinical treatment or follow-up. This worked well for some (mostly mothers); completing the interview while their child was undergoing treatment was a distraction from their many worries and concerns. For others, clinical treatment visits continued to be extremely stressful and they would not consent to be interviewed during that time. Out-of-town/province parents (often just one of the dyad) stayed in Toronto for periods of time when their child was receiving complex care like chemotherapy. For these participants, interviews were planned and completed in a private hospital office and at a time of their choice outside of the daily clinical regimen.

There was an immediate down-side to this whole interview plan. Parents with a child in early treatment were disinclined to participate as they were often struggling to cope moment-to moment with their the new and life-threatening Rb diagnosis. Later in treatment it was often just mothers, or mothers and fathers taking turns, bringing their child in for treatment. The early result was few fathers and long waits for participant-fathers to attend a hospital visit and interview. It became evident that an alternative interview process was required to engage a sufficient number of fathers and dyads to attain viable data collection for father and dyad analysis. First, the researcher offered in-home interviews to study participants living in the Windsor to Kingston corridor in southern Ontario. Second, the data collection time frame was extended to fathers who periodically accompanied their child to Toronto for treatment. Gaps could be months, depending on treatment schedules. Third, was to conduct telephone interviews with consenting individuals residing in northern Ontario and other provinces when no clinical visit or follow-up was scheduled during the extended study period or their adult child was no longer receiving pediatric services at the hospital. This was the least preferred choice but was included and approved in the original REB application. The telephone consent and interview process closely aligned with the in-person practice. Parents expressing interest participating in the study but unable to complete the in-person interview during the study window were sent the telephone information package – invitation letter, telephone interview consent form, and consent to audio-record their session. Mailed packages included an addressed, postage-paid envelop for mail-back of signed consent forms. A follow-up telephone call with each person permitted full review of both consent forms and the opportunity for each parent to ask questions. If individuals decided to continue to participate at that point, they mailed or emailed the two signed consent forms (interview and audio-recording) which I then countersigned and returned a copy for their records. After that step, a time and date suitable to them for the telephone interview. Prior to beginning the telephone interview, I reviewed the signed consents, answered any further questions, and verbally re-confirmed consent to interview and record.

I had met, or had prior face-to-face contact (via projects and clinical observation), with just over one-third of the in-person participants; others were day-of introductions. After formal consents were completed, each interview session began with the introductory topic – a general question on events leading up to their child’s actual diagnosis. Follow-up questions to help frame the topic included: Who helped them understand the disease and risk? and What were their greatest challenges in adapting to a sudden and potentially life-altering event? From prior collaboration, this researcher understood Rb parents really wanted to share their story, to relate individual and family experiences on stress, coping, and ultimate success with a potentially life-threatening disease. Following the instrument skip logic, other sections explored personal demographics, employment and workplace benefits, education attainment/career trajectory, marital status and relationship, social supports, and parenting behaviours.

The only question 40 percent of participants declined to answer was – “Would you be willing to tell me your total annual household income, before taxes, to the nearest \$10,000?”¹² At the end of the interview, each participant was asked if there was anything else they would like to add or if they had any other questions on the project. Participants were reminded that they would receive a written summary of the research project when completed and of the availability of social work support if they experienced any distress after participating in the interview.

In sum, 10 dyads and 2 unmatched parents completed in-home interviews, 10 dyads and 4 unmatched parents completed in-hospital interviews, and 3 dyads plus 1 unmatched parent completed telephone interviews (23 dyads plus 7 unmatched parents; n=53). All types of interviews provided useful information but focus somewhat changed by type. Telephone interviews were the most challenging, certainly for this interviewer but also

¹² Feedback from one examination committee member indicated the passive, open-ended nature of this question invited decline. An active statement on requesting total family income and a card with income response categories would have supported a higher response rate.

for interviewees. Sustaining the information flow was challenging for parents as these telephone interviews averaged one hour. It is important to note 4 of the 7 telephone interviewees resided in northern provincial regions. They provided important perspectives on coping with limited access to follow-up services for adult survivors. One differential from telephone to other interview formats was with the last question. When asked “Is there anything else you would like to add that we haven’t talked about?”m telephone interviewees had nothing further to add or share in the session. Face-to-face (home or hospital) sessions both often produced rich value-added comments not captured in other interview questions, unique parent insights and personal challenges. Parents were most relaxed and participative with in-home interviews. Conducted individually and privately, these were the longest interview times for both mothers and fathers, providing some of the richest information on dyadic stress and coping. The home setting also provided contextual relationship and behaviour/interaction information documented in field notes. In-hospital interviews worked well in all cases. If a parent recognized they were not coping in the moment, the session could be rescheduled. They would return, usually in a few weeks. All these interviews were completed in a private office removed from the clinical areas. For most, interviewing was a break from the surgical waiting room and a chance to tell their personal and family story.

Following recommendations by Patton (2002), the researcher wrote field notes as part of ongoing reflexive practice immediately after each hospital interview or each home visit. This documented important insights, observations, and impressions from each interview in a timely manner. Content was structured around behaviours and descriptions related to respondent expressions and actions associated with the interview content and process. Recording notes and impressions also allowed early thematic insights to emerge.

Coding and data analysis

In a practice sense, recruiting, interviewing, transcribing, coding, conceptualizing, and categorizing interview data overlapped in some fashion for most of the project. The broader process of data analysis actually began before interviews as the researcher spent

considerable time during project development, observing and interacting with clinicians and parents in hospital and external project settings.

All interviews were digitally recorded with an Olympus DS-30 device. Each interview was transcribed verbatim into a Word document using DSS Player Software that enabled easy archiving, editing, and further processing of recordings. Next, the researcher listened to each interview recording while following along by reading the transcript. This was an initial data quality check to ensure that the transcription quality acceptable and a verbatim record of the interview. Corrections were made as required. Then the researcher printed each transcript and hand-coded key themes and key terms/phrases related to research questions and sociological concepts. Interesting and novel parts of each transcript were noted as they may lead to emergent information. As interviews accumulated and it became apparent there were important new themes emerging, the researcher revisited earlier transcripts to confirm and compare specific content. After hand-coding was completed, each transcript was uploaded to Nvivo software where core themes (nodes) had been created. Qualitative coding was based on key terms and themes around disease and demographic variables, sociological concepts, role and dyad experiences – that formed the basis of the interview instrument. The iterative process described for the paper-based coding was replicated in Nvivo. Respondents were tracked and coded as couples whenever possible (e.g., Father 12/ Mother 12). The numeric identifier was assigned on the basis of interview completion. Single parents and those interviewed without the participation of a spouse were coded in the same manner (e.g., Father 7 or Mother 23). Each respondent had a unique identifier for analysis and reporting. The researcher revisited transcripts that had been previously coded, identifying sections that aligned with emerging themes. This process continued until all interviews were completed, transcribed, and coded such that themes were consistently coded across interviews without more emergent information.

Data security and participant anonymity

Immediately after each interview session was completed the compressed WMA file was transferred from the recording device to a password-protected computer in my home office and saved to an encrypted drive partition on the computer. All project files were automatically backed up to an encrypted external storage drive for redundancy. Then the original WMA was deleted from the recording device. All transcribing, coding, and writing was completed on this computer so imported interview files never left the secure environment.

The DS-30 recorder automatically assigned anonymized file names for each session (e.g. DS045). Files were saved and handled with this identifier but the researcher maintained a password-protected Excel log that linked the anonymized file name to the interviewer identity and related contact information. A separate tab also tracked recruitment information – dates, names and contact information, communications, informed consent, and key things to remember about the case. Field notes were transcribed into Word documents and linked to the digital file name. Data will be securely destroyed when all writing and publication is complete.

Ensuring data quality

Qualitative researchers working in an interpretive paradigm must consider data in terms of trustworthiness rather than positivism's criteria of internal and external validity, reliability, and objectivity (Lincoln & Guba, 1985; Denzin & Lincoln, 1994). Lincoln and Guba (2007) identified four essential criteria for ensuring the trustworthiness of qualitative data - credibility, transferability, dependability, and confirmability. Padgett (1998) enumerated six strategies to enhance the rigour of qualitative research - prolonged engagement; triangulation; peer debriefing and support, member checking, negative case analysis, and auditing.

Data credibility

Credibility refers to the degree study findings accurately document and describe participant experiences, i.e. accurately measure what was intended and contextually reiterates those experiences to the reader (Shenton, 2004; Lincoln and Guba, 2007). This researcher employed multiple strategies described by Padgett (1998) to establish credible research. Prolonged engagement with the target population began prior to the formal development of this research through exposure in clinical settings and participation in the National Retinoblastoma Strategy (NRbS). The in-depth semi-structured interview instrument was developed to engage parents in fairly comprehensive conversations that framed their personal experiences in a sociological and theoretical perspective for analysis. Early stages of instrument development included a type of member checking. Four key NRbS stakeholders who were Rb parents completed the interview and provided feedback to ensure questions were easily understood and accurately captured experiences from both the parent and family perspective (Lincoln & Guba, 2007). Peer debriefing and support was used throughout the project. Members of the Retinoblastoma Team provided the researcher with long-term support beginning with a conceptual understanding Retinoblastoma diagnosis and treatment that included situational narratives and contextual observations. Team members provided feedback on the interview instrument and specific clinical information on many cases, before and after parent interviews were completed (often as part of related clinical conversations). Triangulation was essential to the entire research process (Creswell & Miller, 2000; Lincoln and Guba, 1985; Bowen, 2005). To cross-verify the data collected, the researcher reviewed individual case files and spoke with clinicians and social work staff to gain a better understanding of clinical, individual, and family context, both before and after interviews. Attending weekly clinical case reviews provided additional insights on active cases when these parents participated in the study.

Transferability of findings

Transferability is “obtained when findings fit other contexts as judged by the readers or when readers find the report meaningful in regard to their own experience” (Munhall, 2001, p. 300) or demonstrated when “results of the work at hand can be applied to wider population” (Shenton, 2004, p. 69). Transferability of this research was achieved with rich, contextual descriptions and direct quotations to represent participant experiences (Munhall, 2001; Lincoln & Guba, 2007). Results were contextually centred in a Canadian context of care, relevant to treatment centres in other developed countries, but with limited impact to affected individuals and families living in developing countries.

Dependability of findings

Dependability indicates findings remain stable over time; confirmability refers to the internal coherence of the data as in relation to the findings, interpretations, and recommendations (Denzin & Lincoln, 1994). Both criteria are in part established and maintained through an audit trail (Lincoln & Guba, 1985; Padgett, 1998). The project researcher maintained an audit trail of all methods and project decisions so others could repeat the process if necessary (Lincoln & Guba, 2007). Dependability was enhanced through in-person interviews that collected first-hand accounts and perceptions of participants, rather than using secondary sources. Triangulation of multiple data sources for all respondents also enhanced dependability of the research. Confirmability was further supported by the researcher’s ability to practice with consistent objectivity, including designing the instrument, seeking feedback from peers and stakeholders to improve methods and research practices, and following consistent practices to collect data and interpret findings (Shenton, 2004). Triangulation of multiple types and sources of data also enhanced confirmability of the research (Lincoln & Guba, 2007). Finally, a process akin to negative case analysis (Padgett, 1998; Strauss & Corbin, 1990) emerged early in the coding process. Important themes emerged that were not included in the initial coding plan. This required the researcher to iteratively re-examination every case

transcript to assess if emergent themes were applicable. This verification process revised and improved the dependability and consistency of coding across cases.

Chapter 3

Fatherhood role identities and mediators of stress

Cooper's (2000) Traditional, Superdad, and Transitional fatherhood role definitions of modern fatherhood nicely overlay the modern lived experiences of fathers participating in this research study. In no small part, Cooper's definitions are effective here because they incorporate concepts of the new economy and the emerging new definitions of masculinity and fatherhood. Findings in this study aligned with Cooper's original work, showing Transitionals made up the largest proportion of fathers. Further exploration of this category indicated a split into two functional sub-groups which would provide richer contextual analysis. The emergent Transitional-Traditional and Transitional-Superdad subgroups are detailed in the following sections. Of the twenty-four fathers interviewed, coding clearly identified 6 Traditionals, 10 Transitional-Traditionals, 5 Transitional-Superdads, and 3 Superdads. Edley's (2001) interpretive repertoires, ideological dilemmas, and subject positions are used to describe and compare stress and coping strategies/behaviours in each of Cooper's fatherhood definitions, providing a richer context for the lived experiences of these Rb fathers.

Fatherhood roles

Deconstructed using a role-discourse matrix (see Table 1), fathers' narratives revealed the stability of role definitions for some, with dimensions of conflict and the dynamic of transitory or permanent attitudinal and role shifts for others, as they navigated the experiences of their child's Rb diagnosis and treatment. Based on this interview sample, there were no observable generational differences between Traditional, Transitional-Traditional, Transitional-Superdad, or Superdad categories. However, Superdads were 3 to 4 years younger than Traditional fathers at the time of their child's Rb diagnosis.

Traditional fathers

Interpretive repertoires

Traditional Rb fathers used interpretative repertoires embedded in a normative or patriarchal view of family life. As example, Father 16 identified his primary family function as wage earner. He was present for all major Rb treatment decisions for his son but deferred all day-to-day caregiving to his spouse. While Father 16 acknowledged the strain this placed on his spouse, he positioned it as a natural outcome of the necessity of his work. His traditional interpretation of masculinity inherently shaped him to a 'job first' attitude to meet organizational demands that translated into earned income to meet family needs:

I continued to work, [Spouse] was the one who had to be with [Son] all the time and it was tough on her. I was there for the major decisions and referrals or check-ups but the actual treatments and that [Spouse] was there all the time and I could continue to work.

I have to travel, I have to go away and so that ends up being a priority. I don't often have to ... can't say I'm not going somewhere because of the kids or because I've got to work or I've got to travel because I'm the only one working and that becomes what I have to do so that's the unfortunate part and takes me away from the family. I don't have much choice in that one. (Father 16, Traditional)

Even when he was at home, Father 16's personal social and recreational activities took precedence over participating in caregiving:

When I'm home, I try to be with the kids and family but I also try to play hockey and I golf, so also there's some social time away which is healthy but also keeps me away from the family at times too. (Father 16, Traditional)

Mother 16 clearly identified the strains and resentment she experienced from her husband's single-minded focus on work and earnings. Seeing couples bring their child to Rb appointments only enhanced her strain and isolation being the primary caregiver to their affected son:

There was a lot of strain because there's a lot of resentment and like there were a lot of times like he travels a ton and I would see a lot of couples who would go through that together and then it would be like "Well, what do they do for a living then?" that they didn't have to work. There was a lot of resentment. Oh absolutely, it was tough ... there's no question. We don't fight a lot but I harboured a lot of resentment, especially when he could take off and travel and have his dinners out. (Mother 16, Traditional partner)

Ideological dilemma and ambivalence

Conflict often emerges around the caregiving role for these Traditional fathers (Cooper, 2000). They delegated caregiving to spouses, who pushed back by encouraging active participation in parenting and caregiving. Father 24 worked long hours as a senior public administrator. For him, ideological dilemma manifested as ambivalence in multiple life domains – personal, fatherhood, and marital. Asked how his wife helped him to be a better parent, Father 24 said:

So you know she would be ...she'd want to sort of be ... she'd want to unload the kid of course on the weekend too so I would be off with him on sort of weekend outings – him and his friends and that sort of thing. She would encourage me more to talk with him about things more than would my natural inclination..... not that I always followed through on these suggestions. (Father 24, Traditional)

On navigating early fatherhood with a son from an unplanned pregnancy:

Well the whole time was just kind of a troubled time for us on a sort of just the relations between us side of things. But our situation was a ... so it was basically

like what are we going to do you know? It wasn't a, it wasn't clear that ... well I wasn't planning at the time when [Spouse] got pregnant to be living with her, and so ... but we had to cope with it as it evolved. (Father 24, Traditional)

On participating in prenatal classes:

... [W]ell I was going to be there for the birth and so it was just to be supportive and sort of get some picture of what was what was going to happen. It didn't seem to help much when he arrived ... (Father 24, Traditional)

Asked what gets in the way of being the kind of father he wanted to be, Father 24 faulted his own natural reticence to communicate, a deficit he associated with his family of origin:

Oh gee, well probably my own sort of difficulties at communicating would probably be the single biggest thing that gets in the way. I mean I certainly can't see anything in the outside world that's an impediment.

Oh yeah but it's kind of you know.....I don't.....I've never been able to encourage him to sort of talk about feelings, sort of thing that he doesn't like to talk about – I don't want to talk about it myself so I'm not going to.....I've never you know.....I've never been able to sort of coax them out of him so we're kind ofespecially at this stage of his life, it's sort of "How's the day?" you know sort of minimal kind of, kind of answers for the most part. (Father 24, Traditional)

Traditional Father 1 identified a sense of injustice with the Rb experience, interpreting this as a loss of personal power – he could not act to keep his daughter safe. His normative, action-oriented response to this cancer dilemma was experienced in the context of the nuclear family:

Dad's just want to provide and you want to make things right, and you want to fix things, you want to ... and then you can't fix Retinoblastoma, and you can't – you

can't change the outcome of RB – you can't ... so as a dad that's where it hurts. Cause you want to, you want to control, not to be manipulative excuse me, but you want to be able to make things right and you can't. And then I'll tell you that was the biggest cross I had to carry in all this. (Father 1, Traditional)

Subject positions

Traditional fathers used a variety of subject positions from which to view themselves and make sense of their experience. Robb (2004) suggested the most significant other reference male for traditional fathers was their own father. Father 17's narrative referenced his own father when speaking about doing and being a good father. Did he want to be the authority figure, the hegemonic male like his own father, and risk the loss of close emotional ties with his children? Yes meant his children would have similar experiences to his early losses and he did not want that for them. No seemed to imply betrayal of his own father's values and memory. This father coped with his ambivalence by holding a middle ground, offering and experiencing the best of both worlds:

I remember growing up, I remember the relationship I had with my dad. I didn't become friends with him until I was in university. Up 'til then it was kind of like...ohhh, dad, he was the voice of authority and we didn't really talk to him. You know what I mean, you talked to him, but you didn't really talk to him. It wasn't a friendship, it was more of the father ... and university was when I really got to know dad more and it's more ... and so I try to remember that, and try not to have kind of relationship with the kids. Not that it's ... I love my dad and things turned out all right. I think I would have liked to have that relationship as a kid. Obviously, I still have to be the one who ...you know, has to have a firm ... you know, something needs to be done and follow that path, but still I can do it in a way that ... you know what I mean, I don't want them afraid of me and I try to always remember that and do things in a way that the kids are ... you know ... they respect you, they have to ...and they'll listen to you eventually, but it's not a

thing based on fear, it's based on respect. I try to keep that in mind every day and most times I succeed. (Father 17, Traditional)

Superdads

Interpretive repertoires

All Rb fathers/families made use of available health care coverage for treatment, medications, and ancillary care. Many also accessed government and service agency funds for travel and specialized aftercare. Superdads in this study were unique in that their interpretative repertoires were embedded in a care orientation that included attending to emotional, physical, and spiritual needs of those around them. They did not hesitate to use an array of available benefits for self-care in order to effectively participate in emotion work, care work, and paid work. Similar to Cooper's (2000) original findings, the Rb Superdads in this study made full use of available parental leave benefits to actively parent their children while adequately providing for their family:

I was on parental leave for our eldest child and again now with our youngest ... I was pretty much the home dad. (Father 9, Superdad)

With his infant son's sudden and high-risk bilateral diagnosis, Father 9 effectively accessed available benefits in a way that optimized his continuity to do emotion work and care work in the family while maintaining the economic stability of paid work:

I did take time off work, so initially I was on parental leave and I further extended my parental leave and then I was off on S&A (Sick and Accident benefits). It entitles me, to I think, 75% of my gross wages and that's take home so basically, and it's a tax free income ... equivalent to a 40-hour (work) week. (Father 9, Superdad)

Superdads openly acknowledged emotional struggles with their child's diagnosis and actively sought self-care options from medical professionals and available care plans. Father 9 spoke of his personal struggles dealing with his son's complex case of bilateral

Rb with risk of metastases. Holding a broader definition of care that included self, Father 9 accessed supports that allowed him to be present with his family, better able to meet their daily care needs:

I was having a little bit of difficulty dealing with his treatment so my doctor felt prudent that maybe I should take some time off work. I guess technically it would have to be for my own well-being to want to be because I was the one having difficulty. They wouldn't necessarily provide me with the benefits to take care of somebody else, the benefits would have to be strictly to benefit my own well-being so that's what it was about. [Son] was going into his chemotherapy and the bone marrow transplant was coming up so like I said, I was having a little rough time dealing with.... Difficulty dealing with everything so it kind of ... And at that time was the most stressful time leading up to the bone marrow transplant so it was used basically so I could ... I felt obviously better staying at home and caring for my kids and caring for [Son] than I would be at work. So, it made that part of it easier. (Father 9, Superdad)

Ideological dilemma and ambivalence

To the extent captured, Superdad expressions of ideological dilemma and ambivalence were grounded in existential and spiritual justice questions. Father 9's words appear, on the surface, as passive and accepting of the cancer event. In action, this family accepted all care recommendations including a clinical trial. His response is surely contextualized by both cultural influences and location in a multi-generational extended family:

It's really, like his cancer really changed my life. Like I've had uncles, I've had a cousin pass away at a very young ... in his mid-20's of cancer but I mean as a parent, it's just you know, you feel that a great injustice has been done because I mean the circle of life definitely you know the old generations passes before and that's generally the circle of life and when it is broken ... we are trying to stay

positive and he's going to be okay, but he has a serious disease and hopefully he's cured. (Father 9, Superdad)

Cooper (2000) found Superdads sacrificed their personal selves in order to invest heavily in both career and family obligations. That was not apparent through the stories of Superdads in this study, perhaps because they wisely used benefits and family supports to buffer strain. However, Mother 11 aptly described her (Superdad) husband's willingness to accept personal distress to be at the hospital with his spouse for their daughter's EUA:

He's just such a hands-on father, so, I mean, obviously he took two months off work too through the time, but he's never missed ... he missed one appointment like say for this appointment today, he just flew in last night from [Europe]. His dad is in end stage cancer and [expected to die] like today or tomorrow something like that. He came here for you know what I mean, it's just like that's huge – like he's with his daughter for this appointment and when his father is dying like that – to me it's just like “OOOOOH...”. (Mother 11)

Subject positions

Superdads used subject positions infused with an egalitarian gender ideology that includes everyday activities of caregiving:

I try and definitely play with the kids every day and try and instill a little bit of direction so watching them play you know, you kind of like referee and make sure that they play nice and try and help them share and understand and learn things and I pretty much try and do that every day. And at least spend time with them. Definitely, that's the most important thing. (Father 9, Superdad)

Even for Superdads, there is indication of role negotiation with their spouse around 'ownership' of primary caregiving in family life:

Yeah, I think a little more so with like our cooking and primary, at least, I don't know if [Spouse] would agree but I'm primarily the one who cooks and stuff and

I think the way I cook and what I cook I tend to put a little more effort into it and make sure the kids are getting their veggies and things like that so definitely, you know we bought a juicer, we slip broccoli in there so otherwise, I don't think I'd be going to that extent as I do now so. (Father 9, Superdad)

The egalitarian subject position for Superdads is enhanced through strong attachment and interaction with extended family often comprised of multiple cultures and generations:

Family. They have been a big help, that's all. They live in [Ontario], [Europe], all over. I rely heavily on that family support – especially the ones here that they were closer obviously. Yeah, they were very, very helpful on pretty much everything that we needed. (Father 11, Superdad)

We had [Spouse's] mom come in from overseas and then we had my mom who lives here in the city for help so we did manage quite well as far as looking after the kids and when two people were needed at the hospital. (Father 9, Superdad)

Transitional fathers

Transitionals are by far the largest and most varied group of fathers in this study (18 of 24 or 75 percent), a finding similar to that reported by Cooper (2000). Transitionals want to be involved fathers and take responsibility for family caregiving work like Superdads. Yet, like Traditionals they will often marginalize a voiced egalitarian ideology and leave much of the caregiving work to their wives. As a group, Transitional fathers showed diversity in how they contested work and family commitments. In this study, sample size permitted a reclassification of the Traditional group into distinct Transitional-Superdad and Transitional-Traditional categories for enhanced comparison.

Transitional-Superdads

Interpretive repertoires

Transitional-Superdads struggled to translate their aspirational caregiving role into everyday practice because of their strong commitment to work roles. The result was often

child-friendly caregiving arrangements negotiated around work requirements: Father 4, a professional in a large firm with time-sensitive and high-demand needs, described shared caregiving in his Transitional-Superdad fatherhood role. He actively participated in planned and spontaneous activities with his children. However egalitarian the idea of shared caregiving, the time was often negotiated to nights and weekends, based on work demands and deferred to his spouse as required by employer needs:

I spend time with my kids. I came home from work and I didn't make it upstairs to have dinner because my kids wanted to play hockey in the basement. When [Spouse] was with you [interviewer], I know the kids like to go get these slushes so I took them to the store to get slush. I sit outside with them and just enjoy their company, I read books to them at night, they come into my room and we cuddle in the morning ... there's all sorts of things we try to do.

If you sort of divide in half like what percentage I do versus [Spouse], every single weekend I do more than her. Pretty much every minute I'm in the house, I do more. She's here during the day and whenever. I'm here in the mornings or in the evenings or on the weekends, I'm happy to take on and do more than she does.

The only things that get in the way are ... time can get in the way. If I get too busy at work, I try real hard when I'm busy to still come home and still spend time in the evening and if I need to I do work after they go to bed, but sometimes that just can't just can't be avoided. (Father 4, Transitional-Superdad)

Ideological dilemma and ambivalence

Ideological dilemmas were as varied as are Transitional fathers. The unpredictable nature of many cancers induces ambivalence at many levels. The following narratives capture some of the comparators that create dilemma for these fathers.

Father 8 (a Transitional-Superdad) revealed how complex and devastating the Rb experience was for him as a parent. His father died of cancer in his youth. He carries that

loss and now his infant daughter has bilateral Rb with developmental complications. The outcome is far from certain for his daughter. Yet, the Superdad part of him is committed to daily shared caregiving with his spouse, even with a daughter facing life-long challenges including low vision or blindness and developmental delays. Asked what he knew of Rb before his daughter's recent bilateral diagnosis and how he was dealing with the reality, he offered few words for so many unknowns:

Just (what I knew from) from Jeff Healey¹³. It was very difficult. My father died of cancer when I was 16. So yeah, she's so small and fragile. To have that happen to your child is pretty tough. (Father 8, Transitional-Superdad).

Subject positions

Transitional fathers used a variety of subject positions from which to view themselves and make sense of their experience. Transitional-Superdads, like Father 8, frequently defined their fatherhood role by comparing and contrasting themselves with other peer fathers in their neighbourhood:

I'd like to be home more. But I also look around and realize that I'm home a lot. I work 7-3 so I'm home ... I work very close, I ride my bike so I'm home at 3:15 and then especially when the summer comes around, I'll be home for 3-4 hours and see other dads coming home so I feel lucky that you know I have as much time as I do. But, of course you always want more. (Father 8, Transitional-Superdad)

¹³ Jeff Healey was a blind Canadian singer and song writer who was a bilateral Rb survivor. He died of non-Rb secondary cancer as an adult, a common risk for older adult bilateral survivors.

Transitional-Traditionals

Interpretive repertoires

Transitional-Traditional fathers often assumed a limited, but regular caregiving role that worked because it was appended to their work day. Active caregiving for these fathers included negotiating and interpreting their role based on experiences with their own father. Father 22 described his regular weekday routine with caregiving following the evening meal. Activities could be child- or adult-centred and his enacted father role was defined by his childhood ideal of a good father:

I work downtown so for me it's an hour drive. By the time I get out of work and I get here it's 6 or just after. So we have a little time after supper to play, so I try to do something with them or try to involve them in what I am doing, and I try to give them advice. Like, I remember growing up, I remember the relationship I had with my dad ... he was the voice of authority and we didn't really talk to him. So I try to remember that, and try not to have that kind of relationship with the kids. Obviously, I still have to be the one who ...you know, has to have a firm ... you know, something needs to be done and follow that path. I don't want them afraid of me and I try to always remember that and do things in a way that the kids ... you know ... they respect you, they have to ... but it's not a thing based on fear, it's based on respect. (Father 22, Transitional-Traditional)

Ideological dilemma and ambivalence

Transitional-Traditional Father 19's ambivalence stems from self-reflection, comparing himself to his nurturing spouse and a culturally-embedded strong and silent masculine typology:

It's probably my own frailties as a person. My own little things that I don't like about myself. It's probably sitting back and thinking.... Especially with the boys I don't praise as much as I do with my daughters. In some ways, I wish I could be as loving and as nurturing as [Spouse] is. But I feel that in some ways they benefit

from my role as being the ---- it's trying to find the balance between being the loving father and being a positive role model as an independent kind of guy that I want my kids to be. (Father 19, Transitional-Traditional)

In comparison, Transitional-Traditional Father 10's dilemma centred on instilling 'sound' family values in his children when they are faced with competing superficial, cultural messages in public spheres:

It's about trying to build up their own self-esteem really. I'll be honest with her (Rb daughter) at times – hey, not everybody is pretty and not everybody's smart but you are smart, you are pretty but it's not that important either, it's about how you are ... you know ... having good morality in the house for example and morals and then following through on that. They see that, they live that, you know it's part of our foundation then you know you don't have to teach it, it's just there. So hoping that some of that stuff comes through.

But it's still hard to deal with especially in this society which is so superficial I mean she's already hearing that at her age from schoolmates right – one kid said “You're kind of weird, but you're sexy” – 5 years old – there's nothing sexy about a 5-year-old you know, sorry. You can't fight against it but you can sort of work with it to some extent. I guess that's you know really more passive than trying anything ... (Father 10, Transitional-Traditional)

Subject positions

Transitional fathers used a variety of subject positions from which to view themselves and make sense of their experience. Transitional-Superdads, like Father 8, frequently defined their fatherhood role by comparing and contrasting themselves with other peer fathers in their neighbourhood:

I'd like to be home more. But I also look around and realize that I'm home a lot. I work 7-3 so I'm home ... I work very close, I ride my bike so I'm home at 3:15

and then especially when the summer comes around, I'll be home for 3-4 hours and see other dads coming home so I feel lucky that you know I have as much time as I do. But of course you always want more. (Father 8, Transitional-Superdad)

However, Transitional-Traditionals like Father 6, were strongly influenced by normative masculine ideals they encountered during boyhood, including their own father. This shaped Father 6's child-rearing practices, introducing a normative bent that conflicted with his spouse's egalitarian preferences:

My relationship with [Rb Son] is a very physical one. He definitely turns to [Spouse] for the emotional comfort, not so interested in that from me at all. Loves wrestling with me. With ...because he's only got one eye ...without any loss of confidence --- I want him to be fearless. I want him to be a physical being 'cause I think it's important for boys to have that. I was, and I want him to have that and certainly [older Son] has it. So you know I'm always, like if I find myself I'm throwing a ball at him to you know ... learn to catch this, learn to catch it, learn to catch it – figure it out, you've only got one eye you know. And so it's funny, [Spouse] won't allow him to climb up on the windowsill cause she like "Oh, it's dangerous." I'm like ... jump as far as you can – if you bang your head, you bang your head – you'll bounce you know. (Father 6, Transitional-Traditional)

In comparison, Transitional-Traditional Father 5 compared his own childhood activities to that of his son, who is legally blind as a result of bilateral Rb. He recalled playing with friends in his neighbourhood, sees that his son will never have what he considers a normative experience, and struggles with the dilemma of providing a viable alternative that makes sense to him and works for his son:

One of the things sort of I see is that a lot of the kids his age are kind of out and they are all playing and [Son] doesn't seem to really get to do, you know what I mean, to be in the group. I just sort of see him missing that whole you know,

fifteen kids running up and down the neighborhood sort of thing right so I don't know what you are supposed to do and like, he goes swimming and it's a blind swim and he does sort of interact with the other kids but not that much and it's still not the same... like I just remember from when I was a kid you know, I was out the door in the morning and basically came home every so often sort of thing and back out. Whereas I don't think [Son] is going to have that. I just sort of feel that he is missing something there. (Father 5, Transitional-Traditional)

Rb diagnosis was invariably a medical crisis for all participants, with possibility of loss of life and/or increased levels of care over the longer term. In this sample, two Traditional fathers described transformative role identity shifts that emerged several years after their child's treatment began. Individual narratives described a long-term role identity shift, moving away from a Traditional role and towards Transitional-Traditional and Transitional-Superdad roles, that occurred over a number of years and with the supportive intervention of community health professionals. The following description from Father 1 indicates the breadth of role change possible – from Traditional to Transitional-Superdad over an eight-year period:

I, I used to put work before family ... especially in the early years because the overtime paid for a lot of stuff. Now when they call, I just hang up on them basically, because I feel that they robbed me of that time now. My concerns now are “I really don't care” because, you know, I only have them for so long and then they're gone. I don't want to be one of these guys that regrets it and looks back and thinks of that Cat Steven's song you know the cradle and all¹⁴ - that song I'm talking about? I don't want to look back and say “hey I wish I would have done that.” (Father 1, Traditional at diagnosis to Transitional-Superdad after 8 years)

¹⁴ ‘Cats in the Cradle’ was a 1974 Harry Chapin song from his Verities & Balderdash album. <http://www.allmusic.com/album/verities-balderdash-mw0000316279>

Fathers stress and coping

Little sociological research has examined personal stress and coping strategies of fathers as they navigated their young child's life-threatening cancer illness. Limited findings suggest these fathers were uncertain about the future and common coping strategies were to adopt optimistic attitudes and seek information about their child's illness (Mu, Ma, Hwang, & Chao, 2002; Cayse, 1994; Sterken, 1996). This research expands that limited literature, contextualizing in greater detail the stress and coping experience of four categories of Rb fathers participating in this study.

Cumulative stress and stress proliferation

Diagnosis of Rb is a stressful life event for all affected parents and families. Interviewing mothers was contextually very important for this issue. They spoke much more eloquently and descriptively about stress and stress proliferation, perhaps because they carried the bulk of enhanced caregiving, family knowledge mobilization, and interaction with the medical system. Their narratives helped contextualize both father and dyadic stress and chronic caregiving strain when a child has Retinoblastoma. It was common for Rb mothers facing a long, intensive treatment plan for their child to exit the labour force for the private sphere and attenuate related family and dyadic stress. Fathers were not less caring, but they were often experientially more peripheral to the private sphere and less able to describe caregiver role strain. The exception was Superdads who were their spouses equal in providing family care and supporting them at medical appointments, including work leaves for caregiving. Findings suggested both diagnostic factors and disease complexity played a role in parental stress levels and the chronicity of that stress. The impact of a unilateral versus bilateral diagnosis across disease complexity (tumour staging) is detailed in the rest of this section.

Unilateral cases with advanced-stage tumours¹⁵ (D or E) are medically the simplest. Normally enucleated, diagnosis-to-treatment time is quite short and the condition is cured. This life event is an acute stressor with quick resolution and family adaptation as indicated by these two unilateral, enucleation parents:

Father 24 (Traditional) describes the short trajectory for his son:

Yeah, yeah, I mean the treatment was just the enucleation. He was only in there for 2-3 days and then he was, we were, all at home for a week or two. So by the time he started back to school, I went back to work basically. (Father 24)

Mother 25 (unilateral son; traditional full-time caregiving role after treatment; partner not interviewed):

But also we literally took him home after two days. He had a bandage on his eye and I'll never forget friends were coming to visit and family were coming to visit as they were devastated and surprised at this. I was consoling them by that time because we had already lived the 4 days at the hospital and got through it all and you know, found out all we could and were very well educated and already knew what we were dealing with by the time we went home. (Mother 25)

Unilaterals with A to C tumours experience a broader range of therapies and time frames and parents experience higher levels of chronic stress during active treatment. Most initially receive focal therapy (i.e., laser therapy, sometimes augmented with cryotherapy), during regular EUAs occurring over several months. For a small number, in the end the eye is enucleated for cure. Parental stress response varied with the treatment

¹⁵ Rb tumours are staged A to E following the IIRC classification scheme (see Appendix B). Group A are very low risk and Group E are very high risk for metastases. Enucleation for tumour group D & E eyes (high/very high risk) is the primary treatment in unilateral diagnosis.

timeline and complexity comparable to the experiences described in the following bilateral cases.

Mother 27 (Transitional-Traditional partner) described how important that face-to-face conversation with clinicians can be to really understand and accept the chronicity of what they were up against with a bilateral daughter:

One day I was having a really hard time with everything and she [Ophthalmologist] just sat down and said “Look it, it’s going to be bad, probably for 3 years, but she is going to be okay and it’s going to get okay and it will be better but just kind of ride the wave that for 3 years - it’s going to be bad” and so it was kind of like she put me in a state of hearing everything is bad news. She just said “It’s going to be bad for 3 years” so I just remember that day as being okay – it’s going to be bad but then it’s going to be okay. (Mother 27, Transitional-Traditional partner)

Bilateral cases are complex and chemotherapy is often part of regular treatment protocol. Social isolation is a key experience for these families during the active treatment period. However, fathers and mothers describe the experience quite differently. Father 21 (Transitional-Traditional) briefly commented on the strain and social isolation from chemotherapy:

A little bit in that we had to be a little more isolated and certainly through the chemo in the first few years of doing things so you couldn’t go out as much and you couldn’t do things because we had to look after [Son]. So it limited us somewhat sure. (Father 21, Transitional-Traditional)

His spouse, as primary caregiver, had very different yet more eloquently described experience of isolation during their son’s chemotherapy:

It was hard because it was a 3-week period – 2 weeks it was constant laundry because he would be sick. I’m glad he wasn’t on solid food because he would just

vomit constantly. It was crying and vomiting for 2 weeks and then you'd have one week of that and you'd realize that you'd have to go through it again so emotionally it would be awful because you'd be all happy and you'd realize that you'd have to go through this all again. (Mother 21, Transitional-Traditional partner)

Mother 9 (bilateral son, extraocular risk) identified risk of infection inherent in complex chemotherapy/bone marrow transplant therapy for her son and the impact of extended social isolation:

Very significantly because of the bone marrow transplant but first of course when he was neutropenic¹⁶ and was going through chemo we actually had to keep a very low profile anyway. Because we didn't want him to get sick having anything that would delay his chemo, hospitalizations, all of that and then actually after the transplant we spent a fair amount of time in isolation here (at home) as well. (Mother 9, Superdad partner)

Mother 26 (Transitional-Traditional partner) provided a richer contextual description the isolating impact of chemotherapy had on the entire family:

I didn't work after [Son] was born so I was not working for about 3 years. So yes, it had a big impact. Because even when he was home and I decided to do home daycare, kids couldn't come over until he was finished treatment because of risk of infection and that kind of thing. We were sort of surprised at how much you know, the treatment affected everything else. It affected what my daughter could do – she couldn't go to play groups because she might bring home germs. We couldn't have friends over if they might be sick. I couldn't work. So, yeah,

¹⁶ Neutropenia is an abnormally low level of neutrophils, a common type of white blood cell important to fighting off infections, and indicative of infection (Mayo Clinic, 2017)
<http://www.mayoclinic.org/symptoms/neutropenia/basics/definition/sym-20050854>

the fallout was bigger than we kind of thought. We had to be careful of infection beyond the 6 months up to 1-year-old he's still got a blood infection and we believe from the port because he still had the port implanted. (Mother 26, Transitional-Traditional partner)

At the upper end of stress proliferation was Mother 23, a single parent¹⁷ with a two-year-old daughter and an infant bilateral daughter diagnosed at 3 weeks of age. This narrative captured the dynamic and variable nature of bilateral Rb treatment for a child requiring advanced treatment in a tertiary centre:

On treatment stress:

Emotionally, it's the EUAs ... Every time you go in it could be, it's always possible laser, possible cryo, possible this, possible that – and it's never a definite because it's the nature of the disease. You never know what is going to pop up. So that's stressful – you are always kind of on pins and needles while you're waiting ... Every 3-4 weeks. (Mother 23, separated; partner not interviewed)

On travel from another province:

I've like, I've been up for 36 hours. I work as a teacher. I'm a single mom with two kids. So my daughter, my 2-year-old stays home with my sister while I fly to Toronto – it's great, my flights are covered (by a home-province plan) but I pay for any of my accommodations and anything else I spend here. I have to once a month I book 2 days off, luckily I'm with a very big Board [of Education] and they're fairly understanding but I still have to beg and plea for the time and if I go over, I'm going to be docked pay. I still have to do all the work to get a substitute

¹⁷ Father 23 did not interview. Described by his spouse as enacting a Traditional role, he left the marriage and home province while his partner was staying in Toronto for the first 4 months of their daughter's chemotherapy treatment. Father 23 had no contact with his daughters or spouse at time of interview.

in and then worry about my class running while I'm here. So, it's very hard. More stress, absolutely. Right now, it's only missing a couple of days at a time to do EUA's but I was living in Toronto for 4 months during her chemo and I don't know how that would have been had I been working at the time. (Mother 23; on maternity leave at the time)

On out-of-pocket expenses:

If you are doing any sort of chemo, the drug costs whether or not you have any of that coverage. I mean that's just cancer in general ... but they don't again at the beginning they kind of give you a head's up that there might be some kind of cost incurred but you don't know because it depends on the case. And you can't prepare yourself for that. (Mother 23, separated; partner not interviewed)

On stress and social isolation:

Not having enough time, being tired, being exhausted, being stressed out ... Quite often because you want to be strong for your kids your depressing your own emotions, biting your tongue often because you don't necessarily have an outlet who understands. That's the hardest thing ... I have friends, I have family who have kids but nobody has a kid with Rb. It's the most isolating thing ever. (Mother 23, separated; partner not interviewed)

On marital stress related to acute Rb stress:

Like there were issues already, but when you have a sick child somebody kind of has to step forward and really take control and someone falls back and there is resentment. It (separation) was almost right ... even though we didn't call it that, I lived in Toronto by myself (4 months, during chemotherapy for daughter). My mom came from [other Province] and helped me with [older Daughter] from day one. (I) got back from her last three rounds of chemo and [Spouse] had already moved (out of province). (Mother 23, separated; partner not interviewed)

Father 1 (Traditional) described the multifaceted nature and personal, work-related impacts of chronic strain in the early stages of his bilateral daughter's treatment that included socially isolating effects of chemotherapy treatment:

The logistics of family life – of your job, co-workers, people almost getting into your eyes where it's a hindrance instead of a help. You don't want to talk about it every day. Those were the, those were some of the biggest challenges of all. Other than worrying about her health, suffering and that, ... it was keeping your job going, thinking about what you're doing. Like ... I work for [electrical utility], ... so every day I go up [power line poles] and I'm thinking about her. I probably almost killed myself a half dozen times if I didn't have co-workers watching out for me. (Father 1, Traditional)

Spouse as primary mediator of stress for fathers

Brody and Simmons (2007) reported fathers fared best with support from extended family, the church, and health care professionals during their child's cancer journey. This research replicated those findings: these sources of social support were, in various ways, all essential to help Rb fathers and mothers effectively cope. What also emerged was how important the spouse was for the all father's roles identified in this study. Traditional and Transitional-Traditional fathers relied on their spouse as a rudder or stabilizer, keeping them on task and helping them to understand and navigate many aspects of this critical family-life experience. Spousal interaction and activities mediated fathers' stress by providing a framework of supports from where they could focus on work and income for personal and family stability. Father 3 (Transitional-Traditional) relied on his spouse to help him independently research and understand their daughter's diagnosis and treatment. She supported him in his role to lead or co-lead the family through the crisis:

To me I'm more self-sufficient. I had to do it for myself. I didn't need ... I felt like I didn't need experts telling me what was good for my own daughter. So, that

being said, I felt like [Spouse] and I together worked towards that and my own reading and whatever was sufficient. (Father 3, Transitional-Traditional)

His spouse also kept everything running smoothly at home, basically providing him a roadmap to his preferred fatherhood experience:

Because she keeps on top of all these things. She is the one that runs the home because I'm away so much during the day. Her role in the family is completely essential to the day-to-day operations. She's the one who looks after the doctor's appointments, she's the one that looks after the school and making sure that all those things are done. I just show up when I'm told. (Father 3, Transitional-Traditional)

Father 6, a Transitional-Traditional, described the central organizing role his currently at-home spouse provided for his fathering role. She helped him move to a more engaged and involved Transitional fatherhood role by providing information and social connections to map out effective father-son activities that supported continuity of his Transitional fatherhood behaviours:

[Spouse] has read an enormous amount about sort of I don't know about the theory of raising kids but there's been tons of books written about raising boys and raising kids. And so she talks to me about some of the stuff she's read and which hopefully makes me sensitive to some of that stuff. It's a funny question – it's like I'm not even sure I am a good parent ... the part of the division of labour here is I'm at the office a lot and so focused on that part of the life. [Spouse] is currently at home and so she manages sort of the many kids' social activities and extracurricular stuff and in fact actually, so she manages to find programs and things that she thinks I could be part of their lives. For example, every Sunday I take [older Son] to a music class. Now I would never have known, I would never even have come across this class if it wasn't for [Spouse]. So she opens up possibilities and then does some of the logistical legwork to sort of get it going.

Again, through the sort of information network that [Spouse] has – of course, I'm new to Toronto so sort of what's good and what's bad in terms of programs so [Spouse] interacts with all these other moms and they say "Oh, you should check this out or do this" and so [Spouse] brings that information into the household and which I would never be able to do so that's actually a very important role. (Father 6, Transitional-Traditional)

Dependency on their spouse for mobilizing information was also evident in Superdads. Father 9 described how his spouse informs and connects him so he better understood the high-risk nature of his son's diagnosis:

She dives in head first with things and so if something is wrong she is on it right away and she'll jump to the internet and you know, learn everything about it kind of thing so again, translates it for me sort of thing. (Father 9, Superdad)

Traditional and Transitional fathers both held a primary focus on work and work was often a primary source of social support. Superdads were less focused on work and appear to rely more on their spouse for social support and interaction. As for Father 9 (Superdad), talking to his spouse was his primary source of social and emotional support and diversion from the threat of cancer (outside of extended family):

Pretty well on a daily basis, yeah, especially at this time ... it's nice to have ... think about diversions other than you know cancer. It's pretty well at the forefront and it has been for the last couple of years. (Father 9, Superdad)

Mother 9 confirmed the observed Superdad role reversal from Traditionals and Transitionals, putting division of labour in a different, relational context:

We spend a lot of time together – actually more than any other married couple that I know and so it's an everyday occurrence and it's a good thing ... I think in our case at least we make a very, very good team whereas there's a clear division of who is taking what role. I'm the kind of person who really dives in and

research it full force and I mean he's definitely a very, very significant support in that respect. (Mother 9, Superdad partner)

In closing his interview and talking about the intent of this research project, Traditional Father 1 spoke about what he has learned eight years after his daughter's initial diagnosis and how his spouse was the emotional anchor for him during a period of significant stress proliferation:

Dads seem to be outside of the whole picture, like they're the spine of the whole thing, they're the strongest out of the whole relationship. And I don't think that necessarily they are. I think women by design are more stronger to handle stuff like this. That's what I learned from my wife. (Father 1, Traditional)

Coping and social support interventions are key mediators of stress identified in the stress process literature (Pearlin et al., 1981; Pearlin, 1989). This study conceptually and theoretically aligns with Pearlin's model of the stress process. In that context, mothers can be viewed as both agent and conduit of considerable coping and social support resources to fathers. They effectively provide emotional, information, and social supports that mediate stress for their partner and the broader family in this critical life event.

Psychological coping

This next section examines psychological coping strategies of Rb fathers across the four fatherhood role categories. Generally, Traditional fathers coped by orienting around avoidance solutions; Transitionals coped by focusing on problem-oriented solutions; and Superdads coped by enacting emotion-oriented coping from formal sources and problem-oriented coping to resolve the stressor(s). Informal emotion-oriented coping was the domain of Rb mothers, both for delivering and seeking supports.

Problem-oriented coping

Research showed a majority of parents having a child with cancer used problem-focused coping, commonly advocating for their child and actively seeking information about

cancer (Patterson, Holm, & Gurney, 2004). This was also true in this study, as exemplified by Transitional-Traditional Father 6. He used his professional knowledge to understand and contextualize the risk of his son's diagnosis and help him focus discussion with his spouse on the optimal solution. Interestingly, he offered a very normative masculine comparison on the polarizing emotional positioning of fathers and mothers in such a medical crisis, a viewpoint in part likely related to his country and family of origin:

I actually do a fair amount of work in Oncology and the truth is that you know Rb has a 90% cure rate because they enucleate so it's not deadly by any stretch of the imagination. I mean, in fact you know I'd say, it's a cancer that people survive which is rare compared to many of the other cancers that are out there that are obviously more serious. So I think in the back of my mind I never want my son to lose an eye you know that obviously is so obvious but at the same time I knew the worst case scenario was not you know not... it wasn't death. I never felt that I was going to lose him so..... [Spouse] had a much more emotional reaction to it and it's probably pretty common sort of husbands and sort of fathers and mothers. When serious things like that happen, my reaction tends to be let's figure out all the information, let's try to talk sort of the most sensible path to get through this. (Father 6, Transitional-Traditional)

Emotion-oriented coping

Access to both formal and informal sources of this type of coping was essential to Rb families. Mothers in this study were the primary user group of emotion-oriented coping. These interventions helped mothers cope, which in turn meant fathers coped better too. With resistance, a few fathers engaged in formal emotional support (i.e., professional counselling) because the dyad accessed marital services in the community. It may be that the limited engagement with community professionals for emotion-oriented coping supports resulted from ready access to the Rb Team social worker who provided these essential interventions to parents as part of the family-centred model of care.

Formal emotional supports

Superdads readily accessed work benefits to spend significantly more time with families but seeking professional counselling services was identified by only a few couples and some Rb mothers. Mother 1 (Transitional-Traditional spouse) indicated she and her spouse had sessions with a marriage counsellor during the early stages of their bilateral daughter's treatment. She identified a common dynamic that occurs early after diagnosis – overly focusing on the sick child at the expense of other siblings. Earlier conversations with Father 1 told us that was the case for him – he was focused on protecting his daughter and feeling bad that he could not do so in this situation. Spouse 1 experienced his grief as problematic for the family in this situation. Their daughter needed treatment, strategic action was required, and tension caused stress for both of them:

We did seek counseling with regards to our marriage, and something brought forth in the counseling was how we definitely dealt with this differently and [Spouse]'s opinion on how I dealt with it – he had a different opinion than what I think than what it actually was and the reason I say what it actually was because the social worker actually agreed with what I felt with my way of dealing with it. My way of dealing with it was – we have to live for today and I can't sit there and have a pity bath. There was no time for that. You know, there's two other children that aren't part of having to deal with going through chemo or anything and we had to keep moving. (Mother 1, Traditional partner)

Mother 8 (bilateral daughter, complex developmental concerns, Transitional-Superdad Spouse) actively sought out professional support through referral from her family doctor. She described the impact of wait time, her first session experience, and her husband's quite common polite, gendered reaction to an invitation for couple therapy:

I saw, I asked for, I had my physical, my medical physical so I asked my doctor about seeing a psychiatrist. So I saw someone once and I think I've got an appointment next month but holy waiting list – it took like 5 months to go and see

someone – I’m like “are you serious?” but I was just like an hour crying. I’m like “I’m not used to these questions.” And he of course, said “oh, maybe you know, in the future, bring [Spouse].” And [Spouse] is like “oh, boy.” (Mother 8, Transitional-Superdad partner)

Informal emotional supports

Outside of their spouse, Rb fathers did not prefer to engage with informal emotional supports, especially early in their child’s treatment cycle. This was opposite to spousal preference for active peer support from other mothers (and couples) right after diagnosis, and ongoing. Mother 12 was newly immigrated to Canada at the time of their bilateral daughter’s diagnosis. Like other newcomer families seeking Rb treatment, considerable social isolation is experienced and augmented by the absence of proximal extended family. An informal parent peer support network had emerged from the treatment experience of another Rb mother some years prior to match Rb families. This mother recognized that experienced families might be able to offer information and emotional supports to new Rb families with a similar disease profile and expected treatment trajectory. She described the impact of this support:

We didn’t know anybody, actually there was family ... she (Mother 17 in this study), her daughter had Retinoblastoma and (Rb mother), she actually, she took care of us and she help us just to look at my child as a normal child not that you know that she was handicapped. Just to treat her like a normal. She introduced us to another lady and her daughter had Rb as well in both eyes but the eyes were removed and she was, you know, it was showing me that even not having sight you can do so much. (Mother 12, Traditional partner)

Avoidance/denial coping

Avoidance (denial) occurred when the individual used distraction or social diversion to cope with stress. Father 16, a Traditional, described how both a work focus and

additional peer sport and social activities helped him avoid stressors of caregiving during treatment:

I needed a distraction which is work. I also try to play hockey and I golf. Also there's some social time away which is healthy but also keeps me away from the family at times too. (Father 16, Traditional)

Social support

This section contextually explores the extent to which formal and informal modes of social support in each of the emotional, informational, or instrumental categories.

Emotional support

Parent conversations with the treating clinician was seen as the ideal to help them deal with the stress of caregiving (Vanderwerker, Laff, Kadan-Lottick et al., 2005). Limited prior research with Rb parents showed they too preferred direct, emotionally-supportive contact with the specialist treating their child. At the same time, they recognized physician availability was limited and relied on Oncology Nursing Staff and Social Workers as information resources (Fazeli et al., 2006).

Formal emotional support

Formal emotional support in the Rb clinical care setting was primarily delivered by the Rb Team Social Worker. Few fathers and spouses sought out community-based health professionals for formal support.

Informal emotional support

Informal emotional supports were as varied and many as the parents in this study. Sources included co-workers, family and friends, neighbourhood community, church community, and virtual community. Family and friends played a support role for all Rb parents and their families. Traditional, Transitional-Traditional, and Traditional-Superdads most often described ongoing emotional support from a network of close

friends and key family members (often parents and siblings or in-laws). Spouses in general were more out-facing with broader family and community engagement. As discussed, chemotherapy distinctly isolated Rb families from even close friends and family, sometimes for an extended period of time. Superdads and their spouses were much more engaged with familial social support. Mother 11 described the broad social support of an extended family. She also noted the oft-reported experience of being ‘dropped’ by friends and neighbours. Social isolation as a result of chemotherapy was a confounder with some, but not all, of these loss experiences:

I find that well our family is very close in general but since her diagnosis from literally the day she was diagnosed, the family has been like just like by her side you know and I think we had the whole [Ethnic] community came down the day she was diagnosed, everybody was here and it was really great. Friends, neighbours, everybody. I find people want to be a part of it right away and they want to see you, they want to be with you and then everybody disappears except for the family and obviously your closest friends. I found it very weird, like I’ve heard of it before, but it’s just an awful experience when you’re like “Okay, they don’t call me anymore ... that I found when I was here for the two months (daughter’s chemotherapy). My parents came down and I was able to go home for my (university) graduation and some of my closest friends wouldn’t even talk to me. I was just like “what the heck is going on?” They wouldn’t look at me I was like.....I found that very, very weird – it’s very hard. (Mother 11, Superdad partner)

Father 9, a Superdad, described a similar experience with both friends and some key family members:

I did find that with certain friends you know, and some of my family members, some people have failed to That you would normally think would be there as a support network for you have failed miserably. And unfortunately, I’ve had some issues with my sisters and stuff where they’ve just fell flat on their face ... and

then you meet perfect strangers that are neighbours that I haven't even met and they are just there for you one hundred percent. So as far as that goes, ... it was disappointing in some aspects but very reassuring in others. (Father 9, Superdad)

Informational support

Formal information support for Rb parents is consistently delivered by clinical health professionals at consultation appointments and time of treatment, targeted to help them define and understand the problem to improve coping abilities. More is not necessarily better, and Mother 23 described the very common early and frustrating Rb parent experience when encountering both clinical information and narrative:

I only tried pre EUA to do some of the photos, I don't know. I think it would have been better just to have maybe even a diagram I was just seeing my daughter have a speculum put in her eyes screaming like all that was just way too much for me at the get go. And then I had a lot of information being thrown at me – very technical information which I can understand, like I am educated, but I wasn't in the mindset to hear any of that – I just kind of went to see what was going on. And it would have been helpful too I mean people that do drawings and that was the most helpful actually. When they sat down, drew it and said this is where the tumor is stemming from, this is what's happening as opposed to sometimes just showing the photos which to me was not very clear at all. (Mother 23, separated; partner not interviewed)

Father 18, a Transitional-Superdad, also raised the issue of the lack of formal public education around Rb:

I wish that more information was out there. Yeah you have your Art for Eyes but what I know I've only got because we're on the email list. Why isn't there a public information campaign? It's disappointing how that there's not enough information out in the general public about Rb in itself. It's always, if ever spoken ... you know it's a rare disease. Well, it's still a form of cancer and you know

there is always lung cancer, people are aware of colon cancer, all of the skin cancers, there's so many public educational campaigns done for that and I just wish that more would be done to inform parents of the basic stage you know: "Hey you're going to have a kid, these are the things that could happen you know. If you're taking pictures of your child, which you're going to do, take a look, is there a reflection, is there you know all the signs". Do what you can to inform parents and people because you know at our age it's too late. (Father 18, Transitional-Superdad)

In another narrative, Mother 3 (partner not interviewed) showed how informal advice or education can positively impact the psychosocial development of the affected child, his or her peers, and the broader community:

When [Son] was in kindergarten, he made it through the whole year and on the last day of school, his (prosthetic) eye fell out. And it was funny because I told the teachers of course that he had a prosthetic eye and that it could come out and they were horrified. Anyway, they phoned and it was later in the day and he was quite upset ... ran out of the room and didn't want anybody to see him. And so they phoned me to see what they should do. I talked to him on the phone for a little bit ... he wanted me to come in. We had a little talk about you know, this is who he was and this was part of his life and it wasn't going to change no matter what we wanted or wished. It was the way it was going to be and did he want to talk to the kids about it and show them his eye and what made it a special kind of thing. So, I went into the classroom and we had a little show and tell and I didn't go into the whole cancer thing too much. It was just for kids that age, that he had a sick eye when he was born and he had to have it taken out. I focused mostly on the prosthetic because that was cool you know, they make an eye specially for him and it's made out of plastic and they hand paint it. And they got to watch me put it back in. There was only one little girl in the class that didn't want to watch and that was fine. But I heard from other parents 'Oh, my kid came home and now

they wish they had an eye just like [Son]'. So, I think that really helped, at least with the kids in his classroom, to tell them about it and to let them know that it was okay and there was no real big deal. (Mother 3, partner not interviewed)

Instrumental support

Community-based and philanthropic organizations historically and currently provide cancer patients and their families with essential instrumental supports such as financial assistance and respite care that are unavailable from traditional medical sources (Shelby, Taylor, Kerner, & Blum, 2002). Mother 25 relied heavily on the Canadian Cancer Society for instrumental support that covered regular transportation from a more remote northern community as well as ancillary treatment costs not incurred by current parents:

The Canadian Cancer Society was very, very good with me at the time. It wasn't easy ... [Son] was on chemo for about a year and so that meant coming going back and forth to Toronto every 3 weeks. I travelled back and forth faithfully every 3 weeks. They would pay for, I would take a night train and they would pay for my berth – we had... [Son] and I were able to sleep in it and they would pay for my train back and forth and the treatment that [Son] had in Toronto. (Mother 25, parent not interviewed)

Mother 5 had a bilateral son with low vision and relied heavily on Canadian National Institute for the Blind (CNIB) for various social supports to meet her emotional, informational, and instrumental needs:

We do have supports through CNIB and stuff like that that if we have questions "Are we doing this right, is this normal?" whatever with the vision and stuff like that, they're available to us. (Mother 5, Transitional-Traditional partner)

In three instances, instrumental support provided to families through community fundraising was declined by Traditional fathers. Father 7 succinctly expressed the sentiment of this group:

I remember one of our neighbour's girls was going to set up a charity fund for him. I put the kibosh on that because I was too much of a prideful guy. I didn't feel we needed the money ... I didn't want to be perceived as that guy who tried to raise money for a sick kid when, really, we could manage on our own.

(Father 7, Traditional)

Church and community social support

Community can be defined many ways. For this research, community support was experienced by parents through three lenses that are not mutually exclusive – neighbourhood, church, and virtual/online. Churches are traditionally thought of as providing spiritual support for members. This was relevant for two parents:

Mother 24 spoke about returning to active worship to help her deal with her son's questions about death. She identified feelings of greater connectedness and, reflecting back, recognized those supports may have been advantageous when their son underwent treatment in his youth:

It would be interesting too to think about like for us like it did start you know for me at least you know [Son] started asking me about death and you know - did I believe in.....and at the time we weren't really connected to you know the Catholic tradition that we had both been raised in but in fact it's just only very recently that's why we decided to get married that we feel more connected to that and it certainly would have been something that would have helped us at that time. So I'm sure there must be a difference between those who have kind of a spiritual foundation (and those who do not). (Mother 24, Traditional partner)

Traditional Father 1 struggled and spoke of his personal relationship with God as a buffer to his personal questions around why his daughter had Rb and he could not protect her:

Really there are questions that will never be answered and those are the things I deal with ... that's more between Him and I ... you know I've referenced our faith but that's, you know, things are good so I've got no complaints. (Father 1)

Mother 1, Mother 3, and Mother 20 each contextually described broad-based instrumental support many families received from what is often blended church and neighbourhood communities:

Some of the clubs that are attached to the church like the Knights of Columbus, knowing that we were doing these travels, they were doing fundraisers to help pay for some of our travels (Mother 1, Traditional partner)

We had a really supportive community and they did a lot for us while we were gone and you know – we've just always had a very good set of friends. They had fundraisers and raised piles of money for us. You know, [Son] was diagnosed in the spring and we had help with the farm when we were away and people were doing stuff. It sort of showed you who you could really count on. I think it would be more common in small communities when everybody knows your story. (Mother 3, partner not interviewed)

We found out so many people were so supportive and we had a lot of the neighbours and friends from church – a lot of people helped us out even though we didn't have to go through the whole chemo thing and that stuff but we still had a lot of support from everybody. We had meals coming over and stuff for about 3 weeks when we got home from the hospital. We actually had one of our neighbours, the day we came home from the hospital who sent her cleaning lady over and cleaned our house for us. (Mother 20, partner not interviewed)

Virtual communities

Virtual communities are social networks formed or facilitated through electronic media. These social aggregations emerge on the Internet when enough people carry on public

discussions long enough to form personal relationships (Rheingold, 1993; Wellman, 1997; Eysenbach, 2003). Content analysis findings show a preponderance of information and social support, often by sharing personal experiences (Weinberg, Schmale, Uken, & Kessel, 1996). Women were the primary users of these virtual support communities, notable for social support (Klemm, Hurst, Dearholt, & Trone, 1998).

Mother 10 (Transitional-Traditional partner) described reasons for starting a Facebook group that were grounded in a real desire to provide information and informal social support to families so they understood where and how to quickly access proper assessment and care for their child. Some context – diagnosis of her unilateral daughter was delayed because her report of leucorrhoea (the white reflex) in her daughter's eye was, at first, not taken seriously by medical professionals. This delay may have contributed to the tumour staging that meant enucleation rather than more optimal outcomes of early treatment. The couple retained ongoing stress over these events after successful treatment:

I started a Facebook group and that was helpful to me because it has pictures of kids with that white reflex so people can go on there and see. Like it was just such a horrible, horrible week after we found out that I just like I get on there and people who have just got their diagnosis they go on Facebook and do a search and they might find my group and they come on and they say 'We just got diagnosed yesterday or whatever and we're just devastated' and then I can go on there and I can say 'Listen, this is what you need to do or you know. You need to get a hold of your parents, if they're alive, and have them come and take care of your house. You need to send someone to the grocery store and get your meals all lined up. If you have a church, tell them what is going on ... you know what I mean. And then I'll say 'Where you live there's another person on this group so maybe you can message them.' I encourage people if they are having a crappy day – go on and say 'I cannot stand picture day or you know I ran into this woman at the grocery store and she asked me about my kid and you know she was really rude

you know get on and just complain you know'. I'll get on and I'll say 'You know, I'm so annoyed about picture day, I can't stand it'. Picture day is the worst day of my year because it's one day where you just ... there it is you know. (Mother 10, Transitional-Traditional partner)

Mother 11 (Superdad partner) described her affinity for a virtual community such as Facebook as a tool to access short-term, present-moment feedback and social support from other parents of like Rb experience and willing to share online:

Something like that, like I really do – something like that just to hit back ideas and say like “Oh, I’m going down for my EUA and people say “Oh good luck” like they truly understand what you are going through because it’s that waiting really I’d probably say like the day before or maybe two days before the EUA that I find myself getting really like worked up. (Mother 11, Superdad partner)

Employers provided specific mediating supports

Allen’s (2001) work on family-supportive work environments suggested benefit availability alone had only a small effect on favourable employee attitudes to the organization. However, global perceptions of social support from the workplace mapped strongly to employee satisfaction and workplace attachment. While this was not directly a study on employers, current findings map consistently with Allen’s findings and add uniqueness from the father’s role perspective. Of note was the almost universal flexible, instrumental and social support provided by employers. The vast majority of fathers (and working mothers) indicated their employer immediately provided leave, often without them going through a formal approval process. Traditional Father 1 identified social supports from a very understanding employer:

“It was very positive at work. I was off for family reasons and people just picked up the slack and they you know ... very supportive.” (Father 1, Traditional)

Transitional-Traditional Father 6 told how important, and unexpected, the informal social supports from his employer were to him and his family during the intense period of his son's chemotherapy:

I'm lucky, the company that I work for is very tolerant when the shit hits the fan. They are extremely tolerant of people having to deal with personal crises. It's a very... it's a wonderful culture in that regard. The fact that they were very, very cool and supportive. You know, my boss, on days when we'd have chemo, my boss would arrange and she wouldn't even tell me, she'd just arrange to have dinner delivered. And I think in retrospect, something that I was very grateful of that I encountered because I had worked at other places that would have been ... wouldn't have been nearly so understanding so that was very important. (Father 6, Transitional-Traditional)

Father 11 (Superdad) described how his employer handled his daughter's health crisis:

We don't get paid sick leave but the company is being very helpful like the whole time that I've basically been taking off, they are paying me my wages as if I was working steady. We have parental (leave), but I never use it. They didn't even give me that option. They just said take it whatever time you need. (Father 11, Superdad)

Mother 10 (Transitional-Traditional partner) was working full-time when her daughter was diagnosed. She tells a similar employer-support story to Superdad Father 11:

I have family compassionate leave, I have 123 sick days available to me at 75% of my salary, I have 7 (weeks) at 100% of my salary and 4 weeks paid vacation plus Stat holidays and 4 float days. I called my manager on the way back (from hospital) and I said "[Daughter] didn't need glasses, she has eye cancer" and they said "Don't fill out a sheet, just call us when you are coming back." And they paid me all my wages and I accrued benefits and everything, so that's awesome. (Mother 10, Transitional-Traditional partner)

Family culture may impede social support

Superdads and some fathers ascribing to the Transitional-Superdad role consistently offered a narrative involving a perspective on the impact of culture and cultural supports on caregiving stress. Impact was positive in very many of the cases. Father 18 (Transitional-Superdad; spouse is Rb survivor) indicated that family culture (ethnicity) was a barrier to both emotional and informational support for them. Understanding the risk of Rb genetic heritability can only be established with a complete family genetic pedigree tracking back to the proband. This generational profile of a gene-linked condition is necessary to assess and offer genetic counselling on risk of transmission in the immediate and extended family. Identification also allows early medical surveillance and treatment for optimal health and visual outcomes. This was proving difficult in his case:

There is a feeling of resentment – her parents are very closed with information – they don't want to talk to their children about things so (Spouse) hasn't been told a lot about her family history with that and that's very frustrating for me to know that if there is information that they're sticking with the old cultural norm of not giving that information to the kids to protect them. That just bothers me because when it's something this serious, there should be full disclosure. (Father 18, Transitional-Superdad)

Chapter summary

Fatherhood roles

Traditional, Transitional-Traditional, and Transitional-Superdad narratives described aspects of contested masculinity and the differential impacts on fathers' role identities and caregiving behaviours. Superdads emerged as a unique category with a less-gendered, less-conflicted orientation to work and caregiving.

Traditionals enacted interpretive repertoires grounded in ideology-based masculine roles focused on public sphere activities of work and earning. Subject positions were historical,

strongly tied to cultural ideals reflected by their own father. Responsibility for private space and caregiving was deferred to their spouse/partner. Dilemma and ambivalence emerged with expectations of active caregiving and family or dyadic-centred communication. This generally showed in less positive dyadic coping strategies.

Superdads held interpretive repertoires that framed an egalitarian gender ideology and aligned to a private-space and family caregiving orientation; they often assumed the primary role of cooking for the family. Subject positions were not attached to a comparator father ideal or group but shaped or enacted around caregiving and social relationships within culturally and generationally diverse families. Taking parental leave for caregiving was a norm for them. Ideological dilemmas were framed around cancer-centred, existential and spiritual justice questions. Uniquely, Superdads leveraged benefits for self-care and caregiving and work-life balance. These fathers experienced the lowest levels of spousal dyadic conflict around the caregiving burden.

Transitional-Traditionals represented one expression of contested masculinity in this study. Their interpretive repertoire was strongly influenced by both boyhood normative masculine ideals and a positive nurturing role modeled by their partner. Ideological dilemma or ambivalence from this tension was resolved by appending a limited caregiving role to the end of each work day. Family emotional work was often deferred to their partner and the result was dissonance with respect to their egalitarian caregiving ideals.

Transitional-Superdads represented another form of contested masculinity. Their interpretive repertoire included both an active shared caregiving role and a strong commitment to a traditional work/provider role. Subject positions were strongly embedded in a peer father comparator group in their neighbourhood. Dilemma and ambivalence centred on a challenging work-life-family balance achieved by negotiating their private-sphere caregiving role with their partner and around employer time demands. This provided periods of frequent caregiving and engagement with their children but also periods of absence that placed the burden of caregiving on the partner.

Fathers stress and coping

Findings in this chapter show that common Rb-related social stressors are experienced differently across the four fatherhood categories. This in turn can differentiate coping strategies for these fathers. Partners were a primary mediator of stress for fathers in all four role categories. This dependency was especially relevant for Traditional, Transitional-Traditional, and Transitional-Superdads. These fathers actively withdrew from regular family and public-sphere informal peer supports right after the Rb diagnosis, relying heavily on their partner for all informal emotional and informational support for a period of time. Partners/spouses helped fathers in all role identities understand and navigate Rb information so they would be more willing to actively engage in the treatment process. Partners of Transitional-Traditional and Transitional-Superdads organized home life so these fathers could participate in some parenting activities while continuing to focus on work as a providing and coping mechanism. Conversely, Superdads maintained many of their informal peer supports while relying on their partner for additional emotional and informational support. Superdads engaged in a symmetrical or complementary supportive caregiving role at home so spouses could actively seek knowledge, but also other opportunities and activities for the family. Superdads willingly accessed work benefits for extended leaves so they could actively share full caregiving in the home environment.

In general, fathers displayed a wide range of individual coping strategies in this study. Early in treatment, all father types engaged in problem-solving logistical activity in the private sphere, such as organizing instrumental support for travelling to scheduled treatment appointments. Traditional and Transitional-Traditional fathers passed this responsibility on to their spouses as treatment routines stabilized, choosing to refocus on public-sphere work for providing and coping. Problem-oriented coping (i.e., act to alter the stressor) remained a predominant coping strategy for both Transitional-Traditionals and Transitional-Superdads. Avoidance (denial) coping was the domain of Traditional fathers. Work was a needed distraction for these fathers and additional informal social support was provided through peer sport and social activities outside the home. This

category of fathers also uniquely resisted informal instrumental supports (i.e., fundraising) organized by neighbours and church/community members. Emotion-oriented coping (i.e., change your reaction to the stressor) was the domain of mothers in this study, along with Superdads who easily engaged formal sources like counselling to improve coping. Mothers were also the primary users of informal peer social and informational supports provided by other Rb mothers. For some, the connection came through an informal program that matched an 'experienced' family with a 'new' family to provide ongoing support through treatment and follow-up. Increasingly, virtual communities (i.e., Facebook) are the go-to sources of support for Rb mothers, often providing almost instant situational feedback of information and support.

Cumulative stress and stress proliferation was clearly a reality for many Rb families with a bilaterally-affected child. Caregiving needs were much higher in these cases, especially with chemotherapy treatment. Medically-imposed social isolation from chemotherapy protocols challenged fathers and spouses with loss of personal control. Mothers bore the brunt of this socially-isolating care and spoke eloquently of impacts to the entire family. Traditional, Transitional-Traditional, and Transitional-Superdads remained focused on public-sphere work activities. Superdads in the same situation shared heightened caregiving equally with their partner. During these isolating treatment phases, Traditional, Transitional-Traditional, and Transitional-Superdads experienced much higher levels of treatment-induced social stress than their socially-isolated partners or Superdads. For partners/spouses and Superdads, who quite naturally availed themselves of a wider array of formal and informal social supports, engaging in heightened caregiving activities during these intense treatment phases may have attenuated stress. This suggests as a 'caregiving as coping' advantage may be effective in certain contexts.

Chapter 4

Dyadic stress and coping with Retinoblastoma

Across the life course, events, developmental tasks, and daily hassles act as stressors to the individual, the couple, and the family. Critical to this research project was to investigate how couples coped with stressful events related to their child's Rb diagnosis and treatment. The goal was to identify dyadic strategies and coping styles that were effective in managing these acute and chronic stressors and which coping styles had a negative impact on these close relationships.

Research has shown key life stressors can have a significant negative impact on marital satisfaction, spousal communication, and the development/maintenance of close and intimate relationships (Neff & Karney, 2004; Repetti, 1989; Karney, Story, & Bradbury, 2005). Marriages subjected to an overwhelming host of chronic stresses have a higher probability of ending in divorce (Karney, Story, & Bradbury, 2005). This is an important consideration here because anecdotal reports from both clinicians and parents participating in this study indicated the perception of a high rate of divorce among Rb parents. The qualitative nature of this research is not designed to report an accurate divorce rate for these families. However, findings can identify and assess the qualitative impact of multiple risk factors on dyadic coping and marital sustainability.

Systemic-transactional dyadic coping model

Bodenmann's (2005) model differentiated positive and negative approaches using problem-focused and emotion-focused coping strategies. Dimensions of positive dyadic coping included positive supportive dyadic coping, common dyadic coping, and delegated dyadic coping. Dimensions of negative dyadic coping included hostile dyadic coping, ambivalent dyadic coping, and superficial dyadic coping. These positive and negative dimensions are defined below and followed by participant exemplars and contextual discussions. Examples of key mediators external to the dyads (e.g., social and informational supports) follow the discussion of dimensions.

In a medical sense, Rb is cured through enucleation and/or a combination of focal and chemotherapy, supported by bone marrow transplant in rare cases. Cure means life is preserved along with as much functional vision as possible. Impacts of those treatment decisions extend far beyond the clinical setting. Anecdotally, parents have used the term ‘Retinoblastoma family’ to label an identity formed from the experience. Mother 19 (bilateral vision-impaired son; Transitional-Traditional husband) concisely described facets of chronic stress and coping faced by many Rb couples and families as they move to post-treatment living. This narrative also introduced a life course perspective addressed in the next chapter:

It was tough because he was our first child and most of our married life has always had Retinoblastoma in it. It’s a central part of our lives and I find it resurfaces at different times – like sports, school, ... now he’s bringing up driving. So, there are different times when it kind of rears its head. It kind of sometimes brings you back to appreciation, sometimes it brings you back to sadness, sometimes it makes you angry, it’s always kind of there. And your response is different in each situation. We can be watching a hockey game and think “I’m so proud of this kid for how far he’s come and it’s just a blessing”. He’s made that whole scenario and our perception of life, and then yet you still have that grieving and sense of loss. So it’s interesting. (Mother 19, Transitional-Traditional partner)

Positive dyadic coping

The majority of couples participating in this study enacted positive dyadic coping strategies that helped them better understand the disease and treatment process and deal effectively with the shock, loss, and trauma of the entire Rb experience. In many cases, supportive interventions from Rb Team members acted as external mediators or moderators to enhance positive dyadic coping dimensions and/or mediate negative dimensions of dyadic coping. Examples are documented later in this chapter. Three types of positive dyadic coping were documented in this study and are described below.

Positive supportive dyadic coping

Problem-focused positive supportive dyadic coping

Father 9 (Superdad; bilateral son, extraocular risk) described himself as the recipient of positive problem-focused coping support. His spouse acted in a primary knowledge translation role, conducting research to provide practical advice or knowledge that supported his learning. She helped him to orient or reframe to the seriousness of the situation. The result was a mutually supportive coping space, a shared social context, for them to deal with their critically ill child:

She's kind of ... was my go to person as far as information ... she dives in head first with things and so if something is wrong she is on it right away and she'll jump to the internet and you know, learn everything about it kind of thing so again, translates it for me sort of thing. (Father 9, Superdad)

On his spouse's use of internet information to initially identify a serious problem with their son's eye, Father 9 said:

I was a little bit, a little bit doubtful because [Spouse] was the first one to notice the problems and when she first came to me I think ... I came home from work and she told (me) that, she basically said very bluntly that she thought that [Son] has an eye cancer and I thought she was just paranoid ... too much information on the Internet. So I just told her to calm down and you know, everything would be okay. But she was adamant (and) unfortunately she turned out to be correct. (Father 9, Superdad)

Mother 9 confirmed a well-established gender role pattern in the literature, namely that women are the primary user/consumer of health information in families (Nussbaum, 2000; Cotton & Gupta, 2004; Houston & Allison, 2002), using both traditional and supplemental information sources (Pandey, Hart, & Tiwary, 2003). She used this knowledge to advocate for immediate medical care, taking their son directly to the Hospital for Sick Children for care on the following Monday morning:

I'm the kind of person who really dives in and researches it full force and I mean he's [spouse] definitely a very, very significant support in that respect. I was convinced that he [son], well at the very least, that there was something wrong because ... we actually took the photos on a Friday evening so between Friday evening and Saturday I spent probably 4 or 5 hours online researching you know white pupils (Leukocoria), Retinoblastoma, and actually I wasn't surprised by the time of the diagnosis. So we took him to the Sick Kids and he was diagnosed with unilateral Retinoblastoma and his left eye was enucleated. (Mother 9, Superdad partner)

In a different context, Father 29 (Superdad; bilateral infant daughter with developmental issues) described how his spouse acted as the point-of-contact with the medical team. Her role supported him to enter a shared social context, to reframe his understanding of their daughter's disease, enabling a shared sense of "we-ness" that is central to the well-being of these Rb dyads as they move through this life experience:

I remember [Spouse] telling me at some point. I knew about the tumors in her eye. I knew about ... I was at the hospital ... [Spouse] lived at the hospital for a long time and I was there a lot. I remember knowing about the tumors, knowing about everything but it not even clicking that it was cancer. And then one day she told me that oh, the cancer people came by today. Cancer – whose got cancer? Right, I was like oh [Daughter] does. I was just like for a minute, I was like, ... oh yeah, okay. Now it's all kind of coming together. (Father 29)

Transitional-Traditional Father 5 described quite a different problem-solving experience with positive support outcomes. The television show he referenced initially helped him reframe the emerging situation of providing care for a legally blind child. In turn, Father 5 was able to help his spouse reframe the situation to create a common alliance supporting both of them with expectations of life success for their son. This couple described a strong sense of "we-ness" in individual interviews, a sense of mutual trust and commitment that enabled them to successfully navigate this life-changing event.

They also actively used available clinical and community supports to sustain this dyadic alliance:

Dealing with a visually impaired child, you know what I mean, it was nothing I really thought I was going to have to deal with. I do kind of remember sitting in the hospital watching a program and it was on the one blind fellow that climbed Mount Everest and actually watching that show and thinking well, you know what maybe being blind isn't as much as an encumbrance to him as what you know, as what we happen to think it is right? So after that, both my wife and I kind of ... never lifted and laid him and ... we've let him you know, like every once in a while, he's following and tripped over stuff and things like that and you know, it's going to happen. But I'd rather have him outgoing than stand there afraid to move. (Father 5, Transitional-Traditional)

Emotion-focused positive supportive dyadic coping

Father 6 (Transitional-Traditional) described how his wife sought out and co-organized activities that he participated in with their older son. In part, his spouse was at home providing caregiving for their younger bilateral son and Father 6 was the sole income earner for the family. This informal social support had positive emotional outcomes for Father 6, including: attaining the competent and active father role he desired but was not necessarily resourced to do; a sense of belonging through meaningful and enjoyable age-appropriate activities with his son; and an understanding that his spouse believed in his capability as a father. That positive support strengthened a shared social context and enabled mutual feelings of trust and support in the dyad:

Part of the division of labour here is I'm at the office a lot and so focused on that part of the life. [Spouse] is currently at home and so she manages sort of the many kids social activities and extracurricular stuff and in fact actually, so she manages to find programs and things that she thinks I could be part of their lives. For example, every Sunday I take [Son] to a music class. Now I would never have

known, I would never even have come across this class if it wasn't for [Spouse]. So she opens up possibilities and then does some of the logistical legwork to sort of get it going ... I'm coaching under 5 soccer this year. Again through the sort of information network that [Spouse] has of course. I'm new to Toronto so, sort of what's good and what's bad in terms of programs, so [Spouse] interacts with all these other moms and they say "Oh, you should check this out or do this" and so [Spouse] brings that information into the household and, which I would never be able to do, so that's actually a very important role. (Father 6, Transitional-Traditional)

Mother 5 indicated her Transitional-Traditional spouse emotionally supported her on an ongoing basis. As noted in the section above, this couple had an established pattern of "we-ness" enabling a suite of supportive dyadic coping dimensions:

[Spouse] went to every doctor with me because we are just very supportive of each other that way so and we had some scary patches during our pregnancies but I mean [Spouse's] always very good with making sure I was okay emotionally. (Mother 5, Transitional-Traditional)

Common dyadic coping

Common dyadic coping also resulted in the sense of "we-ness" that enabled positive symmetrical or complementary problem-focused or emotion-focused coping, joint information seeking, sharing of feelings, mutual commitment, or relaxing together. This style of coping offered optimal dyad and family outcomes during critical treatment periods and over the longer term.

Problem-focused common dyadic coping

Father 22 (Transitional-Traditional) provided an exemplar of how almost all dyads functionally cope in the acute Rb diagnosis and treatment phase using problem-focused coping to meet the needs of their critically-ill child. As noted in this father's narrative, the

established “we-ness” enabled open and trusted spousal communication that was essential for effective dyadic coping in this critical high-stress situation:

Alright, we need to go to Toronto, we need to get things arranged, we need to go back for the operation ... what day is it? Okay, we'll get set up, I'll be driving, and we just kind of ... I do remember a period where we really cooperated and worked together really well because at that point it was all about [Daughter] and what her needs were. So, and I think ... to me anyways, we've always been pretty good about being able to talk but that was an exceptional time. So, kind of in a bit of a haze, but we just dealt with it and worked together really well. We didn't at all find there was anything that caused us the problem, it was ... you know ... I mean, obviously we were stressed because of the unknowns and that kind of stuff, but we did it together and we didn't ... one or the other didn't get upset with ... about what happened and I do remember that time, we just dealt with it. (Father 22, Transitional-Traditional)

Emotion-focused common dyadic coping

Father 8 (Transitional-Superdad) described the feeling of “we-ness” as a normative experience for this couple. It allowed for a shared social environment where feeling and ideas could be shared with the trusted ‘other’, ensuring each partner’s satisfaction and well-being and in turn the smooth functioning of the dyadic relationship. As well, Father 8 described a key criteria for effectiveness in systemic-transactional dyadic coping which was that each individual in the dyad was attuned to the other’s ‘stress signals’ and were able to offer reciprocal or situational coping support as needed:

[Spouse] is incredible. We're really able to sit down and bounce things off each other and I don't know ... I can recognize when she's under stress and she can do the same for me so we trade off very, very well. (Father 8, Transitional-Superdad)

In a similar fashion, Mother 5 described her experience of this common dyadic coping as “having a natural flow about it”. As noted above, this dyad experienced “we-ness” as a

normative facet of their relationship, one built on past critical life history events often only survivable with mutual trust and commitment. Like Couple 17 (above), Couple 5 effectively read each other's stress signals and interchanged roles in trusted symmetrical or complementary fashion to mediate or moderate emotionally-charged situations:

[Spouse] and I just make a good tag team. I ... we just have a natural flow about it. He can see when I'm at the end of my day and I've just had enough. And he'll step in when appropriate and I can see with him when he needs me to step in, because whatever. [Son] is doing so much better – we've changed his diet and adjusted the medications and stuff but we have, we've had some very rough years with him behaviourally. And it's very exhausting, and every day we're on the phone with the schools working stuff out, not behaviourally, but just, he's very complex. But [Spouse] and I just sit down and say "this is what's going on" and there's never been an issue with that kind of stuff. I think we're the same. We went through ... when we got custody of his son and we went through hell and back, so we've always been good communicators. (Mother 5, Transitional-Traditional partner)

For some Rb dyads, positive supportive coping was situational, enhanced by a shared crisis or trauma. External factors or interventions associated with the Rb team composition in a tertiary care centre often both enhanced positive supportive coping behaviours and moderated existing dimensions of negative dyadic coping. Dyadic impact of these external supports is discussed in a following section. Father 6 (Transitional-Traditional) provided an example of this temporary or situational change, a heightened "we-ness" resulting in mutual trust and commitment in the crisis, then a gradual return to everyday dyadic interaction that contained some level of negative dyadic coping. Father 6 also provided interesting analogy based on his '9-11' (September 11, 2001) experience in New York city:

Temporarily I would say. I think we were more attuned to each other's faults – more tolerant of each other's faults like we were just.... It's funny, in a similar

way I think about, we were in [United States] when 911 happened and one of the phenomena that happened was for about six months ... people were more polite to each other because they had all shared this common tragedy. And then it was ... I remember reading a piece in the [Paper] that said we (are) back to normal because every one's honking their horns again. And there was a sense in which that happened to us to for a period of time we were more ... we more on tip toes around each other. And then it wasn't until we sort of ... it wasn't until we started bickering again that we knew we were back to normal. (Father 6, Transitional-Traditional)

Delegated dyadic coping

The story emerging from narratives in this study suggested Rb mothers acted to solve a dyadic problem and reduce family system stress by stepping out of the labour force for caregiving, at least for a period of time. The result in these families was a shift back to normative tradition, experienced as a 'new' division of labour with fathers primarily focused on the public sphere and mothers primarily focused on the private sphere.

Rb most often occurs in 'new families', young working couples with one or more young children. Treatment, especially for bilateral cases requiring systemic chemotherapy or bone marrow transplant means prolonged periods of intense travel and caregiving for parents. In many of these cases, the mother assumed a full-time caregiver role and the father became the sole income provider. Sometimes that immediate choice was out of caregiving necessity. Longer-term changes in this division of labour were conditioned by the earning power of the father. Where that income is insufficient, mothers often chose home-based self-employment such as child care as a way to continue earning income while fulfilling the primary caregiving role.

Father 16 (Traditional; bilateral son) set the context for an example of delegated dyadic coping. They were a professional working couple with a new home and family. Their first-born son was diagnosed with bilateral Rb and his partner decided to stay at home for the prolonged caregiving period. This couple entered a new, shared social context as a

result of their son's diagnosis. Their decision required mutual goals and a trusted interdependence but it appears the mother's agentic action enabled the new division of labour. To be sure, this executive-father was able to provide adequate income to sustain this new division of labour.

I don't think we thought there was anything abnormal about our first child and getting the house ready and getting our life ready to go and then it threw a little curve at us after that. [Spouse] was working and everything else so she decided to stay home after that. She's a good mother, she's at home. She does everything that needs to be done because I'm away travelling and she's at home and looks after the household and the kids. (Father 16, Traditional)

Father 22 (Transitional-Traditional) described a similar situation and rationale for the move to traditional gender roles. Their young Rb daughter needed a high level of caregiving in the early years and he earned sufficient income to support the family in an ongoing fashion. For Father 22, the new division of labour provided extensive and positive reduction in individual and dyadic stress:

I've been very lucky in that [Spouse] has made the decision, she made the decision to be at home and she just started to go back to work in the last year, a couple of days a week part-time. That first eleven years with her here took the strain off immensely, the worry about having, you know rushing to get them to day care, that kind of stuff. We saw our friends go through that stuff and it wasn't very pretty. It's very tough, tough for everybody and you can luck into the right person to take care of you absolutely, and for some people it has worked out. I think more often than not it's a juggling match that, that leaves everybody a little bit winded in the end, a little bit stressed and we were very lucky that [Spouse] decided to do that and we were able to do that, we were in a financial position that we were able to do that, and that helped immensely. (Father 22, Transitional-Traditional)

Mother 22 described how important the ongoing positive supportive and common coping dimensions were to the decision to become a one-income family. As with Couple 21, the mother's agency appeared to be the driver of role change, choosing to stay at home for caregiving conditional that the husband's income was sufficient for socioeconomic continuity:

He is very supportive and he is always... we made a decision to do things on one income and he was very supportive of that and he helps out with the kids. We get along quite well. We have... he will kind of listen to me when it comes to ... we kind of do my rules for parenting and they seem to work quite well. And really, all three of them are great kids and they are smart. They get good marks and they are well liked by their peers and they have their own little personalities. (Mother 22, Transitional-Traditional partner)

Negative dyadic coping

Negative dyadic coping appeared to be a functional pattern for a few couples in this study, as demonstrated by Father 6's earlier comment: "... it wasn't until we started bickering again that we knew we were back to normal." Considerably more often than positive, these negative dimensions emerged as couples struggled with shock, loss, anger, and blame related to their child's Rb diagnosis and treatment. In most cases, this acute negative dyadic coping diminished as treatment progressed and the case stabilized. Emotional and informational supports from Rb Team members and/or community-based professionals mediated or moderated these negative dimensions and introduced levels and added dimensions of positive coping that strengthened the dyad. The feeling of "we-ness", so important for effective positive dyadic coping, was not enhanced by the three types of negative dyadic coping documented in this study.

Hostile dyadic coping

Father 19 (Transitional-Traditional) described his struggle to cope with his spouse's expressed distress and sense of loss after their son's eye was enucleated. To avoid

acknowledging his own similar feelings, Father 19 minimized his wife's distress and sense of loss by steering the conversation towards a generic problem-solving approach. The result was chronic dyadic anger only resolved through marital counselling. Father 19 also touched on the issue of blame – which one of them was the cause of their child's cancer – a question that emerged as a point of contention for some dyads. The vast majority of Rb cases are not germline, meaning they result from a random genetic mutation and are not transmitted (caused) by one of the parents. This is a difficult fact for some parents to accept:

We were both very angry, we had nobody to angry be with, so sometimes the anger would be directed toward each other – not really realizing it. I can remember one specific instance when we were coming home from the airport. It was when [Son's] eye got enucleated and [Spouse] was saddest because we thought there was a chance we could keep his eye. After the fact, he had very different physical attributes. She couldn't get over the fact, and I noticed it as well, that his eye did not look good and his prosthetic did not look good in his eye and every time you looked at his face, it reminded you that he had Retinoblastoma. And I hear, I am with all my work trying to say “we'll get through this” and [Spouse] kept saying “His eye looks terrible”. I would recognize what she was feeling but I didn't want to go there and I remember having a huge scrap over that one. It took years of counselling, years later we got that one up to really recognize what we were doing. It took two years for the genetic testing to tell us whose gene it was and I'm not even sure to this day which one of us it was. There is no Retinoblastoma in my family. It was actually a random genetic mutation. (Father 19, Transitional-Traditional)

Father 17 (Traditional; adult unilateral daughter) elaborated on the complexity of blame and distancing some dyads experience. Rb disease and treatment information is complex and made even more so with genetic analysis. Even now, some parents receiving genetic counselling for Rb struggle with issues of assigning blame and cause:

After we got through the initial diagnosis, operation and so on we started to learn what is this thing. The Internet was not so prevalent, we did basic homework and again we just found out more about it and tried to figure out where it came from. And that wasn't there were many options. We blamed each other individually. I blamed me because it is hereditary. She said no it's probably me and we are taking this burden on and so on. But it wasn't until years later that we found out that it was neither one of us. (Father 17, Traditional)

Mother 15 (partner not interviewed but was Rb survivor with a germline mutation; unilateral daughter) described the distancing this couple experienced when their daughter was first diagnosed. In context, they knew the risk of transmission to offspring from prior genetic counselling. Still the reality was, for a time, disconcerting. This couple had sufficient positive dyadic coping strengths to effectively communicate during this trying period. Additionally, Father 15 actively shared the treatment journey with his spouse (and daughter), further strengthening the marital dyad:

It was very hard for him because he knew that he ... it's from him that she developed that. I think we went through a period of like, I don't know, separation. That we just both didn't want to deal with it and both dealt with it in our own ways. Then we definitely spent a lot of time together coming to Toronto and talking on five hour trips both ways every two, three and four weeks at the time. That ... I think we spent a lot of time talking about it all. It was a bonding experience. (Mother 15, Transitional-Traditional partner)

Ambivalent dyadic coping

Mother 23 (single parent; bilateral infant daughter) provided the best example ambivalent dyadic coping as her spouse emotionally disengaged and deflected anything to do with his infant daughter's new Rb diagnosis:

Part of the reason we ended up splitting up was because the ... I guess if you want another stress that's a huge stress. Like, there were issues already but when you

have a sick child somebody kind of has to step forward and really take control and someone falls back and there is resentment. There's all kinds of things, but that was kind of the straw that broke the camel's back in our case. He shut down. He stopped talking. I tried to talk about it. I said I think I said something along the lines one day "You know we're a cancer family now". And he said "Don't say that, we're not." I wanted to know everything, you know ... he just passes the buck and ignores. It was always "talk to my wife, I don't know, talk to her, I don't know". He was very happy not to be here. (Mother 23, partner not interviewed)

Mother 17 (Traditional spouse; adult unilateral daughter) described her experience of normative ambivalence inherent in the traditional masculine role with respect to family caregiving:

He (spouse) was always very supportive of my time and energy that I put into the kids. Now having said that, one of my biggest complaints, and I'm sure you know my husband and I are not together, is that I became quite invisible within our marriage. For example, if [Daughter] had an EUA or [Son] had an EUA and I would come home, now looking, back I think he should have had dinner ready and waiting for us or should have made some arrangements that we could have gone out for dinner. And yet, I would come home and find him waiting saying "what are we having for dinner?" And I don't want to blame him because I know I took on the role because what I thought I was supposed to do but we had a very, very clear division of gender-specific roles. And it became quite an issue between us because I could never get him to once they were established to reverse the roles. (Mother 17, Traditional)

Superficial dyadic coping

Father 1 (Traditional) described a dynamic of superficial support that negatively impacted this marital dyad for a long period of time. Actions included judging the other

spouse and offering support without empathy. Later we see positive coping outcomes from effective marital counselling:

You're sharing the same experience but it is different roads even our own marriage. [Spouse] sees things one way and I certainly see things another way. We were very, very close before this happened (daughter's Rb diagnosis), and when it did happen there were times when disagreements, if not borderline arguments, would happen because we'd see things differently. And then it was easier to keep your feelings to yourself than to share them because then it turned into almost a speech ... "Well you need to be tougher", or "you need to be more observant of my feelings". It developed into an argument rather than developing into a support mechanism and so that was something to learn –it was learning how to listen to the other person's views and complaints if you will instead of trying to judge it and that's where we did at the onset I think judged each other. Then we learned afterwards to kind of let it alone which isn't good either but then there's a happy medium in there where we realized it comes down to a matter of respecting each other's view on it and then the support came and then it's been easier.

(Father 1, Traditional)

Mother 10 (Transitional-Traditional partner; unilateral daughter) described her inability to listen and empathize with her spouse in the period soon after her daughter was diagnosed with Rb:

I found I was much more like not respectful. I don't know how to explain it, I might be more distracted in my day-to-day things now, to be able to really sit and listen whatever issue he (spouse) is experiencing. I might be like, "Okay, I got to get the dinner ready and I've got to remember to take my laundry out" (of the washing machine). So he's trying to tell me something and I'm just not all there.

(Mother 10, Transitional-Traditional partner)

External mediators of dyadic coping

The primary focus of this chapter was to document and explore different dimensions of systemic-transactional of dyadic coping (Bodenmann 1995, 1997, 2005) enacted by Rb couples participating in this study. However, these dyads also functionally interacted with an external tertiary care delivery system when their child underwent Rb care, specifically the Retinoblastoma Team and the Eye Clinic in the Department of Ophthalmology at the Hospital for Sick Children in Toronto. Clinical care interactions meant each marital dyad was exposed to a range of information and emotional support, from a variety of team members, staged and delivered to reflect to their child's specific case. These mediating interventions supported or enhanced dimensions of positive dyadic coping and moderated dimensions of negative dyadic coping to help parents optimally navigate their Rb experience.

Father 6 described one of the most important aspects of Rb care for families and fully available only in tertiary treatment centres. The interdisciplinary Rb Team operates on a family-centred model of care to support effective parental decision-making through comprehensive assessment and timely emotional and informational support during treatment and follow-up phases of care. Father 6 also identified a commonly expressed need in the literature, that of parents very much desiring emotional support from the treating physician. However, receiving emotional support is very much dependent on the personality of the physician:

Well, actually, that was one of the things I think was blew me away about Sick Kids. It wasn't ... different individuals gave us different pieces of information. [The Oncologist] obviously was very focused on sort of the course of treatment and the side effects and how are we going to manage the side effects and what we're going to do at home and if what so these kinds of side effects – what are we going to do and who are we going to call and under what circumstances do we go to Emergency and those sorts of things and we did end up going to Emergency a few times just to sort of be on the safe side. And then you know,

[Ophthalmologist 1] was very much sort of the emotional support – that’s sort of her style which I certainly found comforting. [Ophthalmologist 2] seemed be much more “Here are the facts, here’s the data around the disease” ... sort of the more objective, less emotional than many of the physicians in the Eye Clinic you know would ... would sort of give us information about you know the mechanics of the eye and sort of can he see or can he not see and if we lost his eye, it’s not so bad. So what I ... my point I think is that when we ended this process sort of ... of searching for information and answers and what not, Sick Kids sort of ... I really felt surrounded by a team of people who really cared and who knew what they were doing and who were all pulling in the same direction and it was ... It wasn’t so ... it never really felt like an individual. It really felt like a team of people who sort of had been through this before. They knew what was coming. They had a sense of sort of the emotional journey that [Spouse] and I were about to go through. They were sensitive to it, so I mean different individuals provided different pieces of that which I think had it been a single physician it wouldn’t have been anywhere near as ... in sort of a satisfying ... I mean although the outcome was not great (son’s eye was enucleated), I always I do feel that it was a very sort of... I feel very sort of satisfied by the experience. (Father 6, Transitional-Traditional)

Traditional Father 1 provided strong indication how central the Rb Team Clinical Social Worker was for guiding and mediating dyadic communication patterns to reduce negative coping strategies in favour of positive strategies. Effective dyad communication supported couples to talk, listen, and support each other in a brand new health terrain where outcomes were not always certain:

That was set up by Sick Kids because they’re the ones that prepared us that way. And had they not done that, we would have thought probably we were – well we’re not normal or we’re not handling this correctly or we’re not – we’re not allowing ourselves to cry, but they did give us a foundation of it. And the Social

Worker, god shoot me for not remembering her name, she was a Godsend. She shared all that information with us so that's, that's what was able to keep us talking and to identify those conflicts and I hope I'm not getting off topic on it but that's how we've survived so far I think. But that's the stress, that's, that's the stuff that wrecks a family I think. That's where you lose each other and the one important thing that I thought that was very helpful when they came out and said "Here's what we have to offer you, this is what you're going to expect was when they said "8 out of 10 marriages are going to fail when a sick child, when parents have a sick child." And when they told us that, we both looked at each other and made a bond right there. I mean did we have our trials and tribulations, yes, but knowing that right off the get go was a key to making it work if you can believe that. But that was one of the statements that I remember right early on when it came out point blank. (Father 1, Traditional)

Chapter summary

Dyads participating in this study displayed a range of positive and negative dyadic coping dimensions described in the literature. Many engaged in a range of positive coping strategies before their child was ever diagnosed with Rb. A few dyads enabled regular negative dyadic coping in their shared social environment. Rb introduced a new shared social context for which few dyads had effective coping and problem-solving abilities in the initial stages of diagnosis and treatment. There was usually a shift to negative dyadic coping in the period immediately following Rb diagnosis. Supportive Rb Team interventions, delivered in a family-centred model of care and often by a Social Worker, attenuated much of the negative coping related to shock, loss, and anger around diagnosis and social disruption of family life.

Assessing spousal support is key to understanding dyadic coping. This involves capturing process-level information on aspects of the dyadic relationship including the couple's communication practices, challenges around expressing intimacy, parental differences in child rearing practices, and the division of household and child-care tasks. Effective

positive dyadic coping reduces stress for each partner and enhances an individual sense of well-being. This in turn enhances both relationship quality and smooth functioning of the dyadic relationship. The result was low-conflict, high-collaboration dyadic relationships with effective decision-making across all father types, at least during the critical treatment period. Descriptively, these dyads experienced a feeling of “we-ness” – a sense of mutual trust, reliability, commitment, and the perception that the relationship is supportive in difficult circumstances. This change of state occurred in both partners and resulted in symmetrical or complementary positive coping outcomes, in spite of high levels of acute stress. Additionally, some dyads coped by using other types of individual stress management (e.g., work, sports, therapy) and external dyadic support like marital therapy. Superdad dyads were unique in that the couples maintained regular and on-going symmetrical coping behaviours across a range of caregiving and positive support activities in everyday family life, before and after the Rb event. As treatment progressed and life returned to a new normal, Traditional and Transitional-Traditional dyads commonly reverted to a range of positive and negative coping behaviours that were normative in their pre-Rb life. However, a permanent shift to positive, low-conflict, symmetrical coping was experienced by two of these traditionally-oriented dyads. Delegated dyadic coping was a common occurrence in many families dealing with heightened levels of caregiving during treatment. Mothers commonly decided to step out of the labour force to alleviate dyadic and family stress. Only Superdads actively used benefits for extended work leaves to focus on caregiving and spousal support in the home environment.

As noted at the beginning of this chapter, anecdotal clinician reports indicated these stressors were overwhelming and resulted in high divorce rates for these families. Traditional Father 1 recalled he and his spouse were told by Rb clinical staff that “8 out of 10 marriages are going to fail when parents have a sick child.” Study design cannot assess validity or reliability of indicated 80 percent failure rate. In this study one parent from a total of 28 study families (about 4%) reported marital break-up as a direct result of her daughter’s Rb. Contextually, Mother 23 indicated there were already dyadic

relationship problems prior to diagnosis and her infant daughter's bilateral Rb diagnosis was "the straw that broke the camel's back". It seems unlikely that almost all the divorced Rb couples in Canada (of the reported 80%) simply did not participate in this study. Rb parents desire to remain engaged in the dialogue and strongly advocate for an annual Rb research day for Rb families. What can be said with some certainty is that a very large proportion of study couples experienced high levels of acute personal and dyadic stress, especially at diagnosis and during initial treatments. Dyads were challenged to suddenly and effectively communicate and make critical decisions about childhood risks and outcomes that they had never contemplated as new parents. Findings also indicated a strong positive impact of the family-centred model of care in providing effective and timely supports to optimize effective individual and dyadic coping.

A bilateral diagnosis introduced more uncertainty for both clinicians and parents, often leading to prolonged and complex treatment having less certain outcomes and unknown life time risks (e.g., advanced tumour staging in eye(s), secondary risks from chemotherapy, and secondary non-Rb cancer risks in later adulthood). This was experienced as chronic dyadic strain and moderated by both effective social supports and time-dependent factors like treatment success. Situational factors like inter-provincial travel for treatment and self-employment were compounding stressors. Dyadic stress and chronic strain were greatest where Traditional and Transitional-Traditional fatherhood roles intersected with complicated bilateral treatment timelines with sudden and unexpected changes in clinical presentation (e.g., tumour growth reactivated after chemotherapy). In these situations, mothers adjusted their social and marital roles to provide the bulk of child and family caregiving while receiving limited social support from spouses focused on work and earning. The family-centred model of care effectively supports positive dyadic coping strategies for these parents. The interdisciplinary team delivers strategic emotional and informational supports to couples, supporting effective communication and positive dyadic coping strategies. This reduces stress for each partner and enhances relationship quality. The result is spouses supporting each other to stay integrated into their new, shared social experience defined by their child's Rb. This

outcome was evidenced through the many narratives of couples in this study. From a dyadic coping perspective, “we-ness” is at the core of resilience in these dyads as they moved with and through their child’s Retinoblastoma treatment.

Chapter 5

A life course perspective with Retinoblastoma

In this chapter, each of the five life course principles is illustrated with contextual narratives of parent, survivor, and family experiences. Life course principles are often interrelated and commonly appeared together in parent discourse. These interconnections are identified and discussed for each principle. The related constructs of cumulative advantage/disadvantage (CAD) and pathways are documented in a similar manner. CAD is presented first because the concept is contextually present as part of parent narratives in several of the following sections.

Cumulative advantage/disadvantage

Retinoblastoma introduces a wide variety of treatment and outcome trajectories for survivors and their families. The majority of parents with older affected children who participated in this study reported good outcomes for both the child and family. Each journey was unique and not without challenges. Some children were medically more challenged; some families experienced greater levels of acute stress and chronic strain. Much of this reflected the diversity of study participants. These families exhibited resilience and certainly received a variety of social and material support from their families and communities, as well as community agencies and the clinical treatment team. Still there were expressions of cumulative financial disadvantage from a few participants.

Mother 25 (Transitional-Traditional partner), the resident of a northern provincial region, spoke of the longer term financial strain that resulted from travel to Toronto for treatment. The family received travel assistance from the Canadian Cancer Society but there were still lost wages, ancillary costs, and thirty years ago, additional fees for medical services. These costs extended beyond Rb treatment and follow-up; her son also travelled for reconstructive surgery. This family was in the lower third of household incomes reported by participants:

We figure with the (travel and treatment) expenses, with me having to give up all my holidays and time off work and stuff, we figure we put [Son] through university about 3 times. Even with the help of the Cancer Society, it still cost us a bundle. It was a big investment but it's not something you have a choice, it's just something you do. But now when you think back, it was a lot of money - we went through some tough times. (Mother 25, Transitional-Traditional partner)

Mother 28 (Transitional-Superdad) detailed the family's cumulative financial disadvantage that resulted from her son's Retinoblastoma. In context, this was another family in the lower third of reported household incomes in the study. They drove five hours each way to all appointments to save ancillary costs. Mother 28 also gave up her job for five years until her infant son completed active treatment and follow-up. The single income situation further contributed to long-term financial strain:

I guess the major impact now that we are feeling from – the whole thing would be financial. I mean you can only go so many years on one income ... so we went 5 years. So now we are still feeling it. I mean, all the bad credit and all that kind of stuff, it's still affecting us and he (son) is 13 and you think, "Ah, it was like 12 years ago but it's still affecting us that way". You know, we're still surprised how much it still affects you know ... visits, and follow-up, and those kinds of things. (Mother 28, Transitional-Superdad partner)

Geography intersected with income for these two families, creating cumulative disadvantage through increased personal costs of accessing necessary treatment for their child. Social supports and instrumental supports often mediated the impact of stress and chronic strain for these couples. The hospital clinical treatment team provided information and connection to many types of psychosocial and community resources for parents, families, and the affected children. However, study participants residing in smaller and distal Canadian communities often experience greater challenge and more inequity when accessing health care and related support services. In some cases, access to

health care services was constrained because participants were less likely to self-advocate for health resources.

Mother 18, herself a bilateral Rb survivor, spoke at length about the disadvantage she experienced growing up in an immigrant family with parents who wouldn't openly communicate about her very early Rb treatment, the likely family history, or two much-older, pre-deceased siblings. Her story described the intersection of individual, family, history, and culture trajectories to produce early experiences that accrued cumulative social disadvantage into her adult years.

Language and family history were barriers to development of persistent self-advocacy for well-being for Mother 18. Her much older sister always acted as a translator for the parents, including through her Rb treatment. The result was a disconnect from her family and medical history, a sense of early loss. While that did not affect her Rb treatment, the grayness of historical information created a gap, a sense of social exclusion or disadvantage throughout her early years and continued to the current moment. The cumulative social disadvantage of youth transformed into an embodied sense of loss for the adult:

Because of the language barrier and everything I've been told ... I had an older brother and sister that, according to my sister, in her memories and what she can get from my parents, is ... I think they might have had Rb and they both passed away. So again, it's going on my sister's memories because that was in (European country) ... so this is what I'm told and this is what was told to [Ophthalmologist] but I can't say if it's 100 percent. My parents never told me that it was cancer. I remember one day I went to [Ophthalmologist] for my (follow-up) appointment. So when I went in I said "You said that I have a tumour. Is it cancer?" And she said "yeah". So I remember leaving that appointment just feeling like, oh my God, I have cancer.

My parents probably wouldn't have told me it was cancer because they didn't want to scare me. So I think they tried to protect me a lot. And sometimes I think when parents try to protect a child too much looking back now, it's not so good. It's kind of been something that I've struggled with because it was always kind of like "hush, hush, don't talk about it". So I kind of grew up feeling don't talk about it. Don't tell anybody. I don't even I don't know if I've accepted that I'm a cancer survivor. I feel like I never really dealt with it, I never talked about it, I didn't know what was happening to me. Like I don't even know if I realized I only had vision in one eye. I don't know ... I've only ever known to have vision in one eye. I don't know what it feels like to have it in two but sometimes it does feel like when I'm like consciously sitting there I feel like there is something missing here but I have no idea what it feels like. (Mother 18, Transitional-Superdad partner)

Lives and historical times

Rb treatment changed significantly in the last three decades, resulting in a distinct cohort effect for older survivors. Patients 30 years-of-age or older, even those with a unilateral diagnosis, had a greater likelihood of receiving External Beam Radiation¹⁸ (EBR) therapy as regular treatment with complex and advanced Rb tumours. Along with increased risk of secondary non-Rb cancers in adulthood, this cohort of young children receiving EBR for malignant eye tumors frequently developed craniofacial deformities of the skull and eye socket (Jackson et al., 1996). Parent narratives describe the long-term psychosocial impact even after reconstructive surgery on these soft tissue and bone deformations to normalize appearance and facial symmetry.

¹⁸ External Beam Radiation (EBR) therapy directs a beam of high-energy x-rays to the tumor site. Today, EBR is not commonly used as primary treatment modality for Rb and is not recommended for unilateral Rb because of associated long-term morbidity and secondary non-Rb tumour risk (National Retinoblastoma Strategy, 2009: S14, S17).

Mother 25 (partner not interviewed, unilateral son) identified how her son's life course was simultaneously influenced by the historical context of Rb treatment and geographic factors. Medically, her now 30-year-old son was diagnosed as unilateral with advanced tumour staging. He received 21 treatments of EBR plus adjunctive chemotherapy as primary treatment. In the end, his eye was enucleated for cure. This was a normal treatment protocol for the period and disease stage, but long-term impacts for this man and his family are present or still developing. The long term psychosocial impact of facial deformity from EBR is evident, even after reconstructive surgery, and may be experienced as cumulative disadvantage for this individual:

When he went for some of his (craniofacial) surgery with [Reconstructive Surgeon] ... if you have something physical wrong, it doesn't get overlooked in any way, shape or form you know. A physical disability is physical and people, no matter what age, like ... it's just there. You know, our daughter even now says "We go to a bar and people still stare. They are trying to figure out what's wrong." They don't necessarily know what's wrong but they know something is not right. And you know ... and like so, even after all this time, it's still there. (Mother 25, partner not interviewed)

The family resides in a northern provincial region so location limited access to formal and informal peer and professional psychosocial support after active treatment:

[Son] did get bullied and teased at school. We never made a big deal about anything ... we felt he took it well. Probably he ... now in hindsight, I'm not sure that was the best thing. We always felt he was doing really, really well. And really, he did do really well. But in our minds, you know, I don't know if we could have made things easier had he been able to communicate with a professional.

I believe [Son] would have benefitted if he had had contact with other Rb children at a younger age. And we were invited many times over the years – there's been

things – fundraisers and stuff in Toronto but that’s not something we can do.
(Mother 25)

Historical time and geography converged in this case, limiting easy access to Rb genetic and after-care oncology counselling for this family. Several distinct factors influenced the experiences described by Mother 25. First, concerns about current and future reproductive risks and secondary cancers were emerging with her son’s adulthood and greater awareness of disease-related risk. Historically, this was not always effectively communicated to survivors and families and illustrated the challenge of effectively communicating general and specific risk information to adult Rb survivors (see also Mother 30’s narrative on historical practice in the following Section 5.4.1, Within-family linked lives perspective, p. 135-137). Second, thirty years ago genetic analysis and counselling was an emerging science for many diseases, including Rb. This family did not receive today’s level of information because the knowledge was not available for integration into care plans of that time. Third, today’s answers and counselling for this family’s questions were only readily available in a very-distant Toronto:

Our family has not had genetic counselling. Because [Son] only had it in the one eye, I think it wasn’t really pushed at the time. My daughter would really like to have that done. She feels it is important but we haven’t looked into it yet.

I am concerned about [Son’s] health because of the Retinoblastoma. I’m not sure what the research is now, but I believe because he only had it in the one eye his risk of having cancer is pretty much the same as anyone else. However, his boss (Optometrist) ... went into the States and because [Son] had Rb, whether it was one eye or both eyes, his boss came out of that trip saying to [Son] “Your chances of getting another kind of cancer are higher than most people.” I wasn’t there, but he actually took [Son] aside and told him this. And I feel bad for [Son] because now he says okay, what’s the research mom and what should I be doing and stuff like that? He (son) doesn’t have a partner right now but at some point he would like to be married and have children and I’m not sure that, because of the chemo

he was on, I'm not sure that's going to be possible. In fact, 29 years ago that was discussed and then he was on the growth hormone¹⁹ and all kinds of other things.
(Mother 25, partner not interviewed)

A clear knowledge gap shows in this mother's narrative, resulting in confusion and indecision. Mother 25 was correct that her son's unilateral Rb held the population-norm risk of secondary cancer risk. Further genetic testing would simply confirm that. However, as indicated above, her son received 21 radiation treatments and that increased his risk of non-Rb secondary cancer in later life. On that point, the Optometrist was correct but genetic testing would not address that risk. In the absence of information that provided clear context for action, the additional knowledge on secondary cancer risk provided by the Optometrist did not inform how the individual or family act to mediate potential risk. Rather, it introduced a state of chronic strain and worry regarding several very real, interrelated health issues for the son including secondary non-Rb cancer risk and infertility that may have resulted in cumulative disadvantage.

Timing of lives

The timing of an Rb diagnosis differentially impacted the child, the parents, and the dyad in distinct ways, as described by dyad 21 and dyad 24. Rb commonly occurs early, from infancy to three years of age. Couples are starting families and building a life together. Father 21 described the status of family life when their son was diagnosed:

I don't think we thought there was anything abnormal about our first child, and getting the house ready, and getting our life ready to go and then it threw a little

¹⁹ Impaired fertility in both males and females is a major concern for the increasing population of survivors of childhood and young adult cancer. In past treatment, EBR could affect the hypothalamic-pituitary axis to disrupt normal endocrine function balance. Chemotherapy regimens differ widely in their effect on fertility, with some negatively impacting endocrine function over the long term (Wallace, 2011). Growth hormone may be a therapeutic agent for some Rb patients receiving chemotherapy, radiation, or stem cell transplants (Román et al., 1995; National Retinoblastoma Strategy, 2009).

curve at us after that. [Wife] was working and everything else so she decided to stay home after that. (Father 21, Transitional-Traditional)

Mother 21, the primary care giver for their son, provided a much richer contextual description of the increased caregiving strain associated with complex bilateral treatment that included chemotherapy. Here the experience was both parental and dyadic; their infant son was a passive recipient of care at that stage. Family and community were secondary actors in the treatment experience:

He was 3 months old when he was diagnosed. He had that cat eye reflection. First child and the first time around, everyone thought I was nuts. I was lucky because I had a great GP. He called Sick Kids and set up an early appointment. So we went in and [Ophthalmologist] had a look and right away ... It was bilateral Rb. Within 48 hours, he (son) lost the left eye and he still had the tumors in the good eye.

[Son] had 4 rounds of chemo. Apparently it was the short rounds. It still was like an eternity as parents so I can't imagine what it was like if you went through the full 9 rounds. It was hard because it was a 3-week period – 2 weeks it was constant laundry because he would be sick. I'm glad he wasn't on solid food because he would just vomit constantly. He was crying and vomiting for 2 weeks and then you'd have one week off that and you'd realize that you'd have to go through it again. So, emotionally it would be awful because you'd be all happy and you'd realize that you'd have to go through this all again. But we only had 4 rounds of it and I'd tell myself it was only 4 rounds. Imagine if it was double that. (Mother 21, Transitional-Traditional partner)

Today, twelve years later, Mother 21 described dealing with her son's emerging concerns around non-Rb secondary cancer risk faced by all bilateral survivors and their families. His awareness was triggered by the death of Jeff Healey, a bilateral Rb survivor and musician he admired. Understanding and communicating these risks can be challenging.

Mother 21 was excellent at contextualizing the risks for both the reader and her son in an understandable way:

Jeff Healey was a hero – he got to meet him a couple of times and when he passed away that bothered him a lot. Like.... what does that mean for me? Does that mean am I going to die? Those things sit with him and, you know, like I don't want him to live his life I live my life worrying about those things. I don't want them to have to bother him. I just want him to be a normal kid and not worry about those things. We've had the genetic testing and we know what the risks are. He is 50/50 to pass it on. It's up to him if he wants to go back for genetic counselling (as an adult).

I said, when he was worried about the Jeff Healey thing, I said, first of all, Jeff Healey smoked. I said if you smoke, you are going to get lung cancer and that's what he had. Jeff Healey also had radiation. And radiation is huge so I don't know it's not going to turn out. He lost both his eyes and he had a lot of treatments and treatments are different today. You can't compare what happened a generation ago. It's so different. (Mother 21, Transitional-Traditional partner)

Located at the other end of the age-of-diagnosis spectrum, Traditional Father 24 spoke about his 7-year-old son's new Rb diagnosis. Family life and routine was well-established and his son was in grade two. A bee sting near his eye while on vacation initiated a cascade of events that resulted in this late-age unilateral diagnosis. The context around Rb diagnosis for Father 24 was quite different than the infant-timed diagnosis described above by Mother 21. Within the first day parents (dyad 24) needed to inform their son of the impending loss of his eye, a situation not encountered by parents with infant diagnosis. Community was also involved early as their son wanted to immediately tell friends and teachers about having his eye removed because of cancer. This is quite different from the experiences of early-diagnosis parents, who often enter a period of relative social isolation during treatment, before reintegrating into their family and

community. Finally, the evening of that first day brought the serious question – would he be able to see out of his ‘new eye’?:

Yeah, I think [Ophthalmologist] said, well she asked, I’m pretty sure she asked “What do you want to do? How do you want to tell – do you want me to tell or do you want...?” I guess I somehow perhaps stupidly felt I should tell him and of course I’m a dunderhead and just sort of bludged right in and, you know, told him “We’re going to have to take the eye out” and he freaked out a little bit you know. [Wife] promised him ‘Mortal Combat’. We had forbidden violent video games up to that point but that seemed to do the trick. Later that night he, well two things actually, he wanted ... he insisted he had to phone a friend and tell this other little boy who was about a year older than him so 8 or 9 that “hey, they are going to have to have my eye taken out, I’ve got cancer they think.” And this boy’s mother had fatal breast cancer as it turned out and was in remission at that stage but I feel a little bad still that, well, that it actually happened. It was also student teacher night at his school down the road and so we got over fast enough that he went running up to his teacher and said “I’ve got to have my eye taken out” and she of course just was far more freaked out than he was. But then he did ask, I recall ... you know ... it was bath time that night so ... “my new eye, I’ll be able to see out that one won’t I?” That was tough. (Father 24, Traditional)

Mother 24 enriched the narrative around this couples’ navigation of an emotionally challenging situation that threatened the life of their son – telling him he would not see out of his new eye and the use of an incentive for compliance:

I have to say I didn’t really explain it all that well when I explained about having a prosthetic eye because it was really kind of heartbreaking because for some reason he had thought that when he had this replacement eye that he would be able to see again. So that was pretty ... not a very good moment when I had to explain to him that it would just be like a replacement but he wouldn’t be able to see. Well I guess I made it sound sort of ... one of the things I guess when I told

him ... one of the things he always wanted was like to play these kind of combat video games that I never allowed and you know when I told him about the operation, I told him that he could you know, so that was sort of like an incentive for him but you know it's not something I feel great about. (Mother 24, Traditional partner)

Older Rb children can ask existential questions that both challenge parents ability to answer and lead to personal reflection:

[Son] started asking me about death and you know - did I believe in ... and at the time we weren't really connected to the Catholic tradition that we had both been raised in. (It certainly would have been something that would have helped us at that time. (Mother 24)

Mother 24 described the benefits of being actively engaged in the community - social support for her son that came in unexpected ways, including social belonging because of a school yard myth that developed around her son's missing eye:

But actually, one of the most fabulous things was that when he was in elementary school, his gym teacher was great – what she did was she had purchased enough safety glasses for the whole class so whenever they played anything risky it wasn't like he would feel singled out. They all had to wear safety glasses. So that was really amazing and I guess that was sort of special. Oh there were sort of rumors I guess going around the school that he had gotten into a fight but ... I don't think anything really bothered him because he was pretty confident ... I think maybe he even found it a little bit humorous you know. (Mother 24, Traditional partner)

Linked lives

From a linked lives perspective, distinct within-family and between-family differences were observed in parent narratives. Many participants represented new cases with no family history while a few reported multi-generational Rb disease. The within-family linked lives perspective emerged uniquely from narratives of the multi-generational Rb families (see Mother 30 below as exemplar). Heritability of Rb sensitized the family's awareness of disease risk to future generations and they were especially aware of secondary non-Rb cancers in later adulthood. Rightfully, these families put more effort into understanding and mitigating future risks as they, and their future offspring, faced both known and unknown risks associated with Rb. The between-family perspective on linked lives focused on providing peer and family social and informational supports to newly diagnosed Rb families. These social connections, the desire to mentor through shared experience and help new Rb parents understand and navigate the intricacies of treatment, was a stated desire of many parents of both unilateral and bilateral children interviewed for this study. In a sense, an Rb 'community identity' emerged from these formal and informal social support practices and was sustained by annual events. Geography was a limiting factor to participate in these ongoing support activities for many families who received Rb treatment at the Hospital for Sick Children but lived distal from Toronto.

Within-family linked lives perspective

The narrative of Mother 30 (Transitional-Traditional father; unilateral daughter; family history of Rb) exemplified the linking of lives that emerged with the heritable (bilateral) form of Retinoblastoma. First readers are reminded of the impact of history, how medical and risk information was less likely to be communicated to Rb survivors in past generations. This placed risk on future generations when heritable Rb was present in the family. Later in the narrative we glimpse the differential impact of macro change – improvement to Rb treatment; inclusive genetic testing; and the effects that optimal medical surveillance – had on two cousins only a few years apart in age. The older

unilateral female (Mother 30's daughter) was enucleated for cure. The younger, bilateral male was effectively treated without loss of an eye because of early diagnosis and surveillance for all young children in the family. Finally, Mother 30's narrative revealed the complexity occurring with the intergenerational transmission of Rb. The maternal grandmother was bilateral, a fact only discovered after the granddaughter was diagnosed some 45 years after the grandmother was treated. The RB1 gene mutation was historical and indicated a high risk of transmission to offspring. Yet, Rb skipped a generation in this family; three of her four children carried the mutation but none developed Rb. Two of multiple grandchildren developed Rb, one unilateral and one bilateral:

Ours is a very unusual case, my mom was adopted. When she lost her eye in surgery I think it was 1941 or 1942, she wasn't told that it was cancer. She was never told it was Rb so she wasn't able to tell me or any of the rest of us. I have two brothers and a sister. Anyway, the day we got there (to daughter's initial referral to the Hospital for Sick Children), [Ophthalmologist] looked at mom and ... you can see that mom has an artificial eye ... and she said something about, you know, "Oh, you must have had it too". And mom, of course, didn't know what she was talking about. There was a CAT²⁰ scan done on [Daughter] confirming that she did have Rb and they did Cryotherapy every 6 weeks. On the March 28th, [Ophthalmologist] said that the tumor had mushroomed over the end of the optic nerve and so they needed to enucleate, which was done.

She is a unilateral like my mom. We were told it is unusual because of the inherited aspect of it. None of my brothers or sister ever had any tumors. My sister doesn't carry – oh, genetic testing was done of course and my sister doesn't have any mutation. My brothers and I do. Actually, my older brother has two

²⁰ Computed Axial Tomography (CAT) scanning uses computer-processed combinations of many X-ray images taken from different angles to generate a three-dimensional image of the internal spaces of the affected eye. This medical imaging is effective for tumour detection with minimal radiation exposure (Brooks & Di Chiro, 1976).

children who are fine but his third child had it in both eyes. He did not have to have any enucleations though, he was treated differently from [Daughter] ... it was several years after her. (Mother 30, partner not interviewed)

Between-family linked lives perspective

Change often begins with one individual; Mother 17 (now-adult unilateral daughter) described her isolation during her daughter's Rb treatment, the lack of available information and support for Rb, and how her personal actions were the beginning of an informal Rb parent support group:

The dissemination of information was not very good at the time when [Daughter] was diagnosed. I felt like there was no one I could talk to. I called the Cancer Society and they told me they had never heard of a child that young having cancer and they didn't know what Rb was and they could give me the name of a group of kids of parents of children who had leukemia if I wanted to try and get support or talk to other parents ... and I did but that was just totally a different experience. I called the CNIB and said, you know, like my daughter's lost an eye and I need somebody, some help and they said "Well, she's not blind so we don't have anything for you". I went, I searched out and remember back then there was no Internet right, ... so I did get in touch with some parent groups of children with disabilities but it just didn't apply. And we were pretty, I felt we were really isolated. So what ended up happening is that we would go to the EUA's and I'd sit in that room, in the waiting room, and I'd look at the kids with glass eyes and I'd go over and I'd talk to the parents. And I would exchange phone numbers and basically that's how the parents support group started. (Mother 17, Traditional partner)

Mother 30 provided more context to parent group activities (Family Association) – activities, rationale, and outcomes for participants. Here we also see a blending of

generations to tell their story, share their experience, provide social and information supports to new Rb families. The focus was on optimal outcomes, not losses:

We, my brother also helped with the Family Association – he helped with the website for a while, he was the Webmaster. And of course you know that mom was involved. Sometimes at the Symposiums (annual Rb research day for families held in Toronto) we'd be introduced to a family with a little one and I think some of the reason why they liked talking to [Daughter] was that here they could see that she was older and functioning quite well and doing things in her life that parents of those newly diagnosed children questioned whether they'd ever be able to do that. [Daughter] played softball, she has her license, she played musical instruments and did all sorts of things that she wanted to. A few years back ... well, more than a few, I can't remember how many, was it the Discovery Channel did a series on, I think it was 4 Canadian doctors, and one of them was [Ophthalmologist]. Well, our family was in that and they came to the [Home city] Y and filmed [Daughter] swimming to show, I guess, that she can do things that other kids do ... and filmed us here at the house, I think particularly because it is an unusual case and there are generations involved. (Mother 30, partner not interviewed)

Similarly, Mother 12 (new immigrant when her now young adult daughter was diagnosed) spoke of the near and long-term impact this informal peer support had on her family – coping support, reframing away from disability and loss, and paying it forward to the next generation of Rb families:

Naturally, we were actually learning anything about this sickness at hospital. And because we didn't know anybody, actually there was family she, her daughter had Rb (see Mother 17 above) ... she actually she took care of us and she help us just to look at my child as a normal child not that you know that she was handicapped. Just to treat her like normal. And she introduced us another lady and her daughter had Rb as well in both eyes, but the eyes were removed. She was, you know,

showing me that even not having sight you can do so much because the girl she was going by herself to school and she was doing so much by herself.

[Daughter] doesn't look like she has any problems and I think it is very important to keep children in different activities ... and that's why she was in gymnastics and ballet. She had up and down times in ballet but when we started for her private classes she improved much better because she's 1 on 1 and she can see the steps what she has to do and she's dancing really good and she's passing the exams every year and it builds her confidence. I told her "You have to show how you dance for other kids with Rb and just show them that you can dance even when you ... because she's legally blind without one eye but she is doing pirouettes – it's not maybe she can do 2 or 3 times, not 30 times like the other girls but at least she does it. And I told her "You are like model, you have to be proud of yourself what you have achieved" You know what I would like to do for [Daughter]? When we will have Symposium, I will ask to have her solo in front of all the parents with children. It's just 5 minutes, maybe 10 minutes to see that nothing can stop if you want to do it. And she can dance, she can really dance very nice. And I told her "you have to finish the ballet school and the classes because she can get certificate and she can teach for life and you should teach children with Rb". (Mother 12, Traditional partner)

Father 12 (Traditional) had an active leadership role in the national society. He expressed a strong desire to give back through mentoring new Rb families and expressed frustration that these initial connections were difficult to establish:

I'm part of the Retinoblastoma Society ... and I probably could do more as far as promoting this or participating in it. I see that the parents will play such a huge role in getting the best care for their children you know, even in that stage. Can we encourage parents to talk more about this? Probably. (Father 12, Traditional)

I would be very open to maybe consult with somebody who just found out about this ... to share with that very person my experiences that they can actually take and short-cut their learning. That is something that could be beneficial for the parents now. But there is that block ... we never actually had a situation that somebody picked up the phone and called us and said “We understand that you had a close encounter with a disease, can you help me out with how to navigate through this?” We never had a call like this. However, we received quite a bit of help in our early stages because we were exposed, which was a great thing, and thanks to [Ophthalmologist] and Mother 17. I’m grateful. I would like to return that favour to somebody. (Father 12, Traditional)

Human agency

Parent narratives provided examples of human agency for both new-diagnosis Rb parents and adult survivors. Risk-related information provided through genetic counselling was central to empowering informed consent and lifestyle choices as adults. Mother 28 (Transitional-Superdad partner and unilateral Rb survivor; bilateral son) described the impact genetic counselling and early surveillance processes had on personal and dyadic agency and the informed risk to have children when the risk of Rb was present:

We came and talked to [Ophthalmologist] before our (first) daughter was born. [Ophthalmologist] explained all the risks and the plan if anything did happen. [Daughter] came for check-ups frequently. So when we said we were deciding to have another child, [Ophthalmologist] explained the risks were the same as the first child. So we talked about it each pregnancy. We knew as soon as [Son] was born to check for him so he has been under care since he was 9 days old. (Mother 28, Transitional-Superdad partner)

Mother 15 (partner not interviewed but with germline/bilateral mutation; unilateral daughter, twin son with no Rb) provided further insights on increased parental and dyadic agency from genetic testing. The parents accessed genetic testing, were informed of the

risk of germline transmission²¹ from the father, and received counselling on available perinatal management of the disease. The couple chose the recommended amniocentesis²² to inform of Rb presence before birth and early delivery to manage that risk for optimal visual outcomes for the affected child:

[Daughter]'s a special case because she definitely inherited the gene from her dad. We knew that, and she is a twin so we knew too. First we went for genetic counselling and then [Husband] got his DNA map to find out that it was a gene that she could inherit. And then I had amnio testing to find out that one baby had the gene and one baby did not²³. When she was born there was no Rb and then at 3 months they found under anaesthetic that they could see the tumors in the eye. It was very small. She was bilateral. (Mother 15, bilateral partner not interviewed)

Mother 30 offered a different take on agency, describing the couples' conscious choice to have only one child on learning of the hereditary Rb risk after their daughter was diagnosed. In terms of history, their daughter was old enough that they did not have access to advanced testing to further refine understanding of that risk or perinatal disease management techniques available today:

²¹ Germline transmission is the heritable DNA change (i.e. chromosome 13 RB1 mutation) that is carried in the ova or sperm and transmitted to the next generation at conception.

²² Amniocentesis is a procedure in which a sample of amniotic fluid that surrounds and protects a baby during pregnancy is removed from the uterus for genetic testing that can reveal the presence chromosomal abnormalities (Mayo Clinic, 2016). <http://www.mayoclinic.org/tests-procedures/amniocentesis/basics/definition/prc-20014529>

²³ Early prenatal counselling and molecular testing are important for informed family-planning decisions and critical perinatal management of affected babies. When the specific RB1 mutation is known, any at-risk pregnancy can be tested by amniocentesis at any stage of gestation. Parents with known germline mutation (50% risk) commonly chose amniocentesis at 32 weeks' gestation in order to have the option of delivery 2 weeks prematurely at 36 weeks' gestation when there is a 50% risk of a tumour being present (National Retinoblastoma Strategy, 2009, S21).

We only have one child; that was a conscious decision – not because we didn't want to have to deal with a child who had Rb but remember how I said I always was concerned about what she was experiencing at the moment and based on the fact that ours is a hereditary type of Rb and that normally it's bilateral and I think given that ours was inherited and what it normally is, we got off lightly (unilateral diagnosis). (Mother 30, partner not interviewed)

These narratives showed that effectively communicating risk to patients is essential for informed agentic action on the part of the patient and/or their caregivers. Yet, for some parents, understanding and acting on cancer risks and uncertainties was because they really did not understand the language of risk (concepts like odds or likelihood). Mother 15 provided a real example of the 'elasticity' of parent thinking with respect to understanding likelihood of Rb occurring in their child. In spite of accessing genetic counselling and testing for definitive information on her daughter's condition, she held the expectation a shorter treatment pathway with reduced risk was possible, a position that conflicted with clinical reality:

We knew the possibility but you don't listen though. It was really hard for me because they told us that there was a 90% chance that the child who was carrying the gene would develop it but I just thought for sure we'd be in the 10% zone. It was hard news to hear however. Like you know, there are many sick people out there and I knew that if our child is to be sick, that's what she is going to have. That was okay but still you are very much in shock. My husband is bilateral too. He has one eye. Instead of thinking that she was more apt to have a more serious case of Rb, I thought the reverse. I thought because [Husband] was 28 at the time, and I thought 28 years of medicine ahead of us, I thought she would be like one smooth appointment and a little treatment and done. So I was in shock to know what was in store for us. However, [Husband] did lose an eye to it so I still..... I thought Tory would never have to go through that because we did everything right in our hopes and dreams kind of thing. After the first meeting, I understood

how serious it was. The first time they came out of the operating room and told us we had to be back in 2 weeks and it was going to be a long stretch of 2 to 3 years. (Mother 15, bilateral partner not interviewed)

This narrative showed how important context can be in decision making. Mother 15 held a personal perspective that discounted risk because of anticipated advances in medicine over the quarter century. Formal education alone was not likely a factor here as she had attained a postsecondary credential.

Life-span development

Parent stories in this and previous chapters revealed the uniqueness of individual biographies and the diversity of life patterns in human development in the presence of Retinoblastoma. Rb acted as a biological change agent in these families, impacting individual life-span development in various degrees from the formative years forward. There is also evidence for considering life-span development at the level of dyad and family as indicated in work by Bengtson & Allen (2009).

The greatest life-span development challenges and changes occurred with bilateral Rb presentations. This heritable form can significantly impact multiple aspects of individual biographies over the life course. Bilateral patients often have greater treatment risks (e.g., chemotherapy), greater likelihood of at least partial vision loss in saved eyes, risk of secondary cancer in later adulthood, and risk of disease transmission to future generations. These factors in turn induced stress and coping challenges for the individuals as they matured, but also for parents, the dyad, and sometimes the wider family.

Individual-level developmental change

Mother 14 (Transitional-Traditional spouse) described how her son was diagnosed as bilateral at three months after being examined for a lazy eye. Treatment was one enucleation, with chemotherapy and focal therapy on the remaining eye. Parents receive comprehensive genetic counselling at treatment that includes managing future risks, including use of protective eye wear when playing sports and future risks of smoking.

That said, ongoing challenges are often experienced and reinforce the importance of communicating that risk in an understandable and actionable form. As for many parents, managing their child's individual life-span development is challenged with the arrival of adolescence and the ascendant importance of peer socialization. Prescribed risk management protocols were out the window with this young man as individual behavioural and social factors began to potentially reshape future developmental trajectories:

He was very good about wearing it (safety glasses) until high school. That was the end of it. He was very active in sports playing basketball and volleyball and he refused to wear his glasses ... and it didn't matter what we did we just couldn't force him to wear them. His social life is a problem. He drinks excessively (and) ... the other day he had a cigarette ... [H]e's so intelligent in school but he can make really stupid social decisions in life. He knows he's prone to cancer and it (smoking) could cause cancer. He's already got the mutation in every cell. He knows that, he knows smoking could do that. (Mother 14, partner not interviewed)

A risk counselling session with the Oncologist proved ineffective too:

He's an adolescent and he's impatient ... he would lose focus and wasn't listening to her. (Mother 14, partner not interviewed)

A second example of individual developmental change was described in the prior section examining fatherhood role definitions. Two fathers described transformational role changes that developed over time and as a result of personal, dyadic, and family factors related to their child's Rb. Fathers interviewed for this study were located in one of four role definitions, each having fairly unique performance parameters. All these fathers were able to initiate temporary changes in state/role to deal with the medical crisis of Rb. Permanently shifting dimensions of self (e.g., interpretive repertoire and subject position) to improve personal and family coping was outside a normative model of fatherhood role

development. Developmental shifts for these fathers were associated with, and perhaps dependent on, other changes within the dyad and family.

Family-level developmental change

Findings in this chapter suggest dyads were similar to fathers in that they could increase functional or positive coping behaviours to deal with the Rb crisis. Similar to individual fathers, most dyads returned to a normative pre-Rb relationship and coping dynamic as their child's health improved. Often supported by Retinoblastoma Team intervention, dyads enacted emotional or problem-solving positive supportive coping approaches to support each other and effectively deal with their child's illness. Many of these dyads described heightened symmetrical coping behaviours during that period. Superdad dyads were unique in that symmetrical coping or "we-ness" was a core component of the everyday dyadic relationship prior to their child's Rb. Some dyads maintained that developmental change to positive symmetrical coping over the longer term. That shift specifically co-occurred with those fathers permanently shifting their role identity from Traditional and towards Transitional-Traditional or Transitional-Superdad definitions.

Disease-treatment matrix shapes life course experience

In the context of Rb, life course pathways are co-shaped by both the formal delivery of care model and the presence of active genetic eye disease. Hierarchical referral pathways and standards of practice 'stream' new families into care based on diagnostic criteria and disease complexity. Rb is clinically described as unilateral or bilateral presentation. However, those labels do not capture the essence of the disease-treatment matrix. When genetics are taken into account, affected individuals either do or do not have the mutated RB1 gene and that differentiates the treatment pathway; parent experience and social trajectories; and potential outcomes over the life course. As a result, not all cases are treated at a tertiary Rb centre like the Hospital for Sick Children. Many unilateral cases receive full treatment and follow-up at secondary Rb centres in other Canadian cities. The down side with this model is that secondary treatment centres do not all have the full multidisciplinary complement to provide often essential social supports to families.

Individuals not carrying the RB1 mutation

The primary treatment for unilateral Rb with advanced tumour staging is enucleation of the affected eye. Father 24 described the treatment ‘simplicity’ of his son’s advanced-stage unilateral enucleation:

Yeah, yeah, I mean the treatment was just the enucleation. He was only in there for 2-3 days and then he was we were all at home for a week or two. So by the time he started back to school, I went back to work basically. (Father 24, Traditional)

Sometimes the unilateral diagnosis is early with small tumour staging. In most of these presentations, the child would receive focal therapy for cure. Recurrent and/or new tumours in the eye can be expected so the surveillance and treatment timeline can extend months to a few years. Adjunctive chemotherapy could also be used in the treatment plan. Mother 28 described her unilateral daughter’s treatment timeline that included complex therapy to save her eye and vision:

It takes about 4 or 5 hours for us to drive here. When we were first diagnosed, we weren’t allowed to go home for I think two months ... two and a half months. And then we were coming every 4 weeks for a year and now it is every 6-8 weeks. It’s been 2 years. She had chemo, the cryotherapy, the focal therapy – no enucleation. They said she is only the third or fourth child that they have been able to save the eye. It’s easier to tackle because now, like she’s pretty much on the homeward stretch. (Mother 28, Transitional-Superdad partner)

Individuals carrying the RB1 mutation

Mother 5 described the essence and uncertainty of the bilateral (RB1) experience. Her infant bilateral son presented with what might be termed a worst-case scenario – the loss of both eyes and blindness. Yet, the outcome was different than expected, not an uncommon clinical experience with this disease. The large tumour in her son’s remaining

eye suddenly calcified (stopped growing), saving partial vision. This was a big gain for the entire family but only the beginning of the life course journey for son and family:

They (Ophthalmologists) told us ... what they actually said was that the lazy eye was actually ... had died. It wasn't viable and it had to come out. The other eye they said it was a very large tumor – they were going to give us time to mourn over the one eye before they took the second eye. But then remarkably [Son]'s tumor calcified very quickly ... everything balanced out and he was actually able to keep that eye because originally they didn't think so. So, we went from thinking he was going to be completely blind to where now he has some perception of light and shapes of objects and stuff like that. (Mother 5, Transitional-Traditional partner)

Mother 30 (Transitional-Traditional father; unilateral daughter; heritable Rb) described the core intergenerational and life course challenges of secondary non-Rb cancers that may affect families with the hereditary mutation. Her mother lost an eye to Rb as did her daughter. Experientially, it was as if nothing had changed in 45 years. As well, the affected generations in this family had aged sufficiently so the concern around secondary cancer was a manifest reality for the grandmother and remained a future possibility for her daughter at the same time. Mother 30 was also affected, but was not herself a Retinoblastoma survivor:

I think the concern over what medical future she (daughter) has is always there, especially considering the fact that my mom had a secondary (non-Rb cancer) and now what I'm looking at with myself (personal new cancer diagnosis).
(Mother 30, partner not interviewed)

Chapter summary

The diagnosis of Retinoblastoma is a sudden transition to a new life course trajectory for the entire family. New and unique stressors, the emergence of sudden and interdependent

role changes, and changing social trajectories influence and shape parent role identity, coping, and caregiving.

The younger Rb cohort no longer faces social disadvantage of facial disfigurement from radiation treatment. However cumulative disadvantage factors persisted for some affected families. Resource-based disadvantage occurred when lower family income was linked with Rb mothers taking extended (multi-year) leave from the work place for caregiving. The chronic financial strain persisted years after active treatment was complete. Ease of access to essential and follow-up health care services was reduced for families having to travel from northern and out-of-province locations for specialized Rb treatment. In spite of available external resources, financial and emotional strain was compounded for lower-income families.

A life course perspective is directly affected by personal/family elements of biography, history, and social structure. The Rb trajectory is further affected by a disease-treatment matrix that differentially streams patients for medical care. Unilaterals typically receive care at a secondary treatment centre closest to home. Treatment is simpler overall and families are not subjected to prolonged social and relationship disruption. The more complex unilaterals and all bilateral children are treated in tertiary care in Toronto. Delivery of complex care is lengthy with heightened stress and coping challenges for parents. This treatment-related stress is compounded with heightened caregiving needs for affected children. Those travelling far and/or staying for prolonged periods of treatment also experienced chronic social disruption in many life areas and high levels of personal and dyadic stress.

A unique perspective on within-family and between-family linked lives emerged for those bilateral families in this study. Within-family linkages focused on the understanding and mitigating risk associated with secondary non-Rb cancers of later adulthood and intergenerational transmission of Rb to future off-spring. Understanding genetics through access to ongoing counselling was an important coping factor. Between-

family linkages focused on peer mentoring to contextually provide informal social and informational support to improve coping in newer Rb families.

Finally, the dynamic nature of the life course perspective suggests developmental change can occur at any point in a trajectory. Two fathers in this study permanently shifted from Traditional role identities to Transitional-Traditional and Transitional-Superdad role identities. These transitions occurred over a period of several years and change was augmented by the fathers' willingness to engage with professional community-based individual or dyadic counselling support during that period.

Child psychosocial oncology research on father and dyadic stress and coping is largely absent of a sociological perspective. Retinoblastoma research has primarily focused on medical and genetic aspects of disease and care, along with some work on effective knowledge translation to support fully-informed consent and effective parent decision-making. The result is an inherent knowledge gap that minimizes the sociological understanding of stress as it relates to father and dyadic experiences, challenges, and necessary supports required for optimal coping during the highly challenging period of diagnosis and active treatment for a young child's Rb. This qualitative study applied sociological lenses of fatherhood role identity and discourse, the stress process, and the life course perspective to address five research hypotheses related to father and dyadic stress and coping when a child was diagnosed and treated for Retinoblastoma. Research discussed in the previous chapters strongly indicates that valid and meaningful answers to these hypotheses is conditional on three criteria: the complexity of the disease presentation in each case, the role identity held by each father, and the dimensions of dyadic coping at play in each relationship. The five research hypotheses are addressed as part of an integrative discussion in the next chapter.

Chapter 6

Discussion

Retinoblastoma (Rb) is a medically complex genetic eye cancer of infants and young children. As with other childhood cancers, Rb treatment can be long and intensive with uncertain outcomes. Treating and managing the disease can be challenging enough for medical experts. However, Retinoblastoma does not occur independent of parents, families, and unique social worlds. Rb parents – individually and as dyads – are exposed to significant, often chronic stress just when new social and parental roles are actively being negotiated in both private and public spheres of life. Clinical disease management takes on an entirely new context – helping parents understand and effectively cope with sudden and often life changing events for which there is little advanced warning.

Sociological research on fathers' social stress and coping and dyadic coping is largely absent from child psychosocial oncology research. This study begins to address that gap, using the Stress Process Model to examine fathers' social stress and coping, the Systemic-transactional Model to examine dyadic coping, and the Life Course Perspective to examine stress and coping over longer trajectories related specifically to bilateral Rb. Most life course concerns described in this study were expressed by mothers and were related to future Rb-related risks to their adult child and her/his offspring. Mothers identified and described these risks/concerns far more effectively than fathers in this study. This heightened awareness to well-being may result from developmental awareness acquired from both their role as a primary caregiver and their regular contact with medical professionals for the broader well-being of their child(ren). It's not that fathers don't care about their child's social and emotional well-being. It appears from their narratives that they locate large aspects of their identity and activity in the public sphere where education and work and organizational performance are primary measures of success. Their bilateral child's future success in these domains were one of the main concerns identified by these fathers. Dyadic stress did not appear to be enhanced from

this role-based division of concern. This may simply reflect acceptance of normative gender roles in the dyad. However, many of these dyads had early and extended exposure to a clinical team that supported positive dyadic coping strategies as a key component of clinical disease management. Perhaps these early interventions influenced the developmental trajectory of these dyads towards higher overall coping. There is opportunity for future research here to explore long-term coping outcomes that emerge as a result of supporting positive dyadic coping strategies at diagnosis and throughout lengthy treatment periods.

For this comparative research, fathers were placed in one of four functional or descriptive role identity categories. Classification was based on key elements of their identity discourse identified as they described themselves at the time their child was diagnosed. Findings suggest fathers strongly identify with or ‘live’ one of these four role identities in daily life. However, role adherence could be conditionally flexible and resulted in a range of situationally-focused behavioural sets for fathers in this study. Both the normative or ‘lived role’ and ‘conditional role’ co-occurred with and impacted dyadic coping. These identity roles could differentially influence dyadic coping, conditional on how these fathers coped with personal social stressors.

In critical or high-need situations like the diagnosis of Rb, almost all Traditional and Transitional-Traditional fathers shifted to a Transitional-Superdad role with a heightened caregiving role and enhanced common or positive dyadic coping. This role-based shift to egalitarian caregiving and dyadic support was most often temporary, with most father types reverting to their normative pre-Rb role as their child’s treatment plan was established and their partner assumed the expanded caregiving role at home. Fatherhood role and dyadic transitions were permanent in three instances in this study. In one dyad, described by Mother 23 as having pre-existing high levels of ambivalent dyadic coping, sudden marital separation and full disengagement by the father occurred shortly after diagnosis. By all accounts, this fathers’ role transition and resulting trajectory change for himself and the rest of the family was permanent. In two other cases, Traditional fathers

experienced a permanent role identity shift – one to a Transitional-Traditional role and one to a Transitional-Superdad identity. These were not transitions in the life course context. Rather, these changes are suggestive of psychosocial maturation processes reflecting substantial and permanent identity and relationship change that occurred over an extended period of time and with supportive intervention of professional counselling services. The two associated dyads also reflected heightened common and positive dyadic coping and an increased sense of personal and dyadic well-being by the end of that period of each father's developmental change.

The findings around role transition is suggestive of a conceptual developmental model for fathers' role identity, one that impacts dyadic coping and may have value for improving the current model of care for children with Rb and their families. The observed shift from Traditional to a Transitional-Traditional or Transitional-Superdad role is complex to frame from these descriptive categories. However, certain contextual factors may point to a structure for future explanatory research. The two Traditional fathers above shifted both discourse and role identity. Identity shift was most obvious as they moved from deferred caregiving and limited emotional support in the private sphere to negotiating a more active caregiving and supportive role with their partner. Discourse shifted on multiple fronts – work remained a core identity/activity and caregiving commitment was organized around employer needs. However, public-sphere activity and an idealized masculine role was no longer the primary source of identity and coping for these fathers. The Transitional-Traditional father acknowledged affinity for his partner's nurturing role that strongly contested his identity reference to the masculine ideal of his own father. This buffering resulted in a regular but limited caregiving role in evenings and on weekends. The Transitional-Superdad changed his reference from a cultural masculine ideal embodied by his own father to peer fathers living in the neighbourhood. This emergent masculine reference group is unique in that normative cultural ideals are replaced by a relational reference group of fathers with similar demographics and exhibiting a range of similar active caregiving and private-sphere activities. Conjointly,

these dyadic relationships shifted towards positive and symmetrical coping that provided both members a heightened sense of trust, belonging, and well-being.

Other co-factors seemed to influence these developmental role identity shifts. First was time - these fatherhood identity shifts occurred over several years after their child was diagnosed. This suggests an active psychosocial maturation process might be at play, perhaps beginning even before the Rb diagnosis. These dyads experienced differing and conflicting personal coping strategies in crisis; negative dyadic coping was the early the norm. In both cases, dyads regularly engaged with the Retinoblastoma Team Social Worker to improve immediate coping. They also sought out community-based professional marital and personal counselling services for further support. In sum, individual parents and dyads cope optimally with critical life events like Rb when both are engaged/engage early in regular and ongoing positive dyadic support.

Encouraging/motivating fathers to regularly attend all treatment sessions with their partner and child may be an effective first step to improve immediate and long-term well-being for Rb fathers and families.

The optimal end-state for individual father, dyadic coping, and family well-being may indeed be the Superdad role identity. It is not clear from this descriptive research how that final role transition to Superdad might happen in the nascent developmental model proposed in the prior paragraph. That gap itself may provide insights towards thought on an explanatory developmental model of father role identity. Considered in an explanatory context, Superdads in this study exhibited a unique cluster of family, cultural, and skill/training precursors that were collectively absent in the lives of those in other fatherhood role categories. Superdads were located in multicultural and multigenerational families living in Canada and Western European countries. This possible socialization influence coincided with a less-gendered, egalitarian fatherhood role identity enacting high levels of active caregiving in the home and the extended family.

Dyad coping was positive, symmetrical, and egalitarian. It might be argued the unique family dynamic shaped role identity for these fathers. What's confounding is that all

these Superdads each held certification as a journeyman in a skilled trade. This seems paradoxical within a historical masculinities perspective where blue-collar work is aligned with hegemonic expressions of masculinity. However, skilled trades emerged from an artisanal history where masculinity was not judged solely on physical brawn. For these skilled workers, masculine identity was not just subjective, competitive, and role-based as in modern organizations. Perhaps like artisans of old, these Superdad journeymen defined their workplace masculinity objectively (or less subjectively) through the outcome, the end product, of their skilled work. If that supposition is true, Superdads might frame their active caregiving role from an equally objective perspective. Effective caregiving requires a different set of acquired skills and the outcome is well-being of family members.

Chapter 3 closed with a suggestion that Superdads' engagement in active caregiving in the home attenuated high levels of personal, treatment-induced social stress and might be viewed as a 'caregiving as coping' advantage. An addendum could be that the artisanal training of Superdads 'allows' them to hold objective definitions of caregiving (the product of their active caregiving work) so they enact differing, even contested, fatherhood identities as required and without dissonance. Additional support for this conjecture comes from the two Traditional fathers who permanently shifted role identity to Transitional-Traditional and Transitional-Superdad identities. Each also held journeyman status in a skilled trade and their family histories traced to Western Europe. However, these were not the dynamically engaged multicultural and multigenerational families of Superdads. This is suggestive that a family socialization model may be a partial explanation for Superdad role identity.

There may be an emergent 'process versus outcome' approach here offering some explanation as to why some fathers were able to change role identity over time and many others enacted a single role. Reiterating from above, fathers holding journeyman status may frame their masculine role in terms of an objective outcome or end-product of their skilled work. This provides a strategy for adaptive role identity (i.e., home versus work)

without dissonance. Fathers with organizational roles aligned to management and professions – most fathers in this study – are much more likely to experience their masculinity as a subjective, competitive, role-based identity. In this study, the Traditional fathers not holding skilled trade certification were professional business and finance managers in larger organizations. Transitional-Traditional and Transitional-Superdad fathers also held a variety of professional and managerial positions in organizations of various sizes. Knowledge worker roles are commonly exposed to ongoing performance evaluation processes with outcomes that are not always objective. It may be that these fathers have their role identity defined by subjective process measures embedded in organizational culture. If that is the case, masculine role identity for these workers may be experienced as a recursive process shaped by organizational practice. Minimal skill-based outcome role identity is possible in this context. Regular performance feedback provides a narrow set of subjective measures from which a ‘successful’ masculine role can be defined and enacted. Organizationally, fathers adhere to a single role identity that fits, and is located in, the public sphere. This becomes the default fatherhood role identity as there is no other avenue for feedback to establish a solid alternate role identity. Traditional fathers hold a role identity located in the public sphere role and minimize their private sphere activity as coping. Transitional-Traditionals and Transitional-Superdads were identified as expressing contested masculinities. What may be happening for these fathers is the strong recursive maintenance of a work-based role identity contests their aspirational desires to be a participatory father and partner. Resolution of these contested roles is strategic for fathers holding either of these role identities. They uniquely establish a workable work-life role balance that is supported by or negotiated with their partner. The subjective/process versus objective outcome explanation of fatherhood role identities offered here requires further conceptual and theoretical work before explanatory research is possible.

The next section provides a detailed summary for each of the five research hypotheses. The impact of fatherhood role identity on social and dyadic coping is contextually

explained for each hypothesis. Dyadic coping and life course considerations are discussed.

Hypotheses

Hypothesis 1: The relative absence of fathers from the medical domain during the early discovery and treatment phase results in increased personal and marital stressors.

This hypothesis is conditionally true, based on fatherhood role identity at diagnosis and existing dyadic coping strategies. One dyad experienced high levels of ambivalent coping before their daughter's bilateral diagnosis and marital separation occurred shortly after treatment began. The father was not present at clinical diagnosis, fully disengaged from initial treatment planning, and left the family shortly after treatment began. Other study fathers actively participated in home/dyadic life at the time of diagnosis and all shared the discovery/diagnosis experience with their partner. They attended initial medical appointments and were active participants in instrumental actions to access optimal care for affected child. Differentiation began with the active treatment phase: Traditional and Transitional-Traditional fathers quickly reoriented to the public sphere, returning to a traditional work and earning role. Of note, these fathers rejoined their partner in the medical process for any secondary critical treatment decisions. They did not use work benefits to attend medical appointments with partner and child. These two father types shifted additional burden of care, including responsibility for medical appointments, to their spouse very early in treatment. Traditional fathers experienced the least overall situational stress because they focused on public-sphere activities including peer social activities. Their avoidant coping strategies increased chronic strain for their partner, in part because of higher caregiving responsibilities and low levels of informal social support. These dyads also exhibited ongoing negative dyadic coping but remained functional in all respects. Like Traditionals, Transitional-Traditional fathers focused primarily on public-sphere work activities during regular day time hours. They provided active caregiving that was limited to weekday evenings and on weekends. This regular

private-sphere caregiving and informal social support reduced partner role strain and positively impacted dyadic coping. Transitional-Superdads were most conflicted with respect balancing public and private spheres. They aspired to shared caregiving with their spouse while remaining strongly attached to their work role. Generally strong positive dyadic coping strategies permitted negotiation of shared caregiving to fit work demands and minimize caregiver stress for both individuals. Transitional-Superdads accessed work benefits to regularly accompany their partner and child to all medical appointments during active Rb treatment and follow-up care. Superdads were unique in that they chose to access work benefits to move from the public sphere to the private sphere to provide active shared caregiving with their partner. They also used work benefits for formal emotional support from community health professionals. They reported the lowest levels of personal and dyadic stress, even when family caregiving needs were high. Uniquely, Superdads remained engaged with peer and family members throughout their child's Rb experience, receiving and providing a range of social support. The outcome was optimal ongoing well-being for themselves as well as partner, family, and extended family. Superdad dyads held a strong sense of "we-ness"; they enacted various positive dyadic coping strategies and continuously collaborated for optimal outcomes for all members of the family.

Hypothesis 2: Role strain persists for fathers as treatment outcomes for their child challenge traditional beliefs and activities expected from the father-child relationship.

This hypothesis was true for Traditional and Transitional-Traditional fathers who described this type of role strain when treatment outcomes meant reduced vision and physical limitation for sons. They held a normative expectation that sons should be socialized in part through 'rugged' play and sports, much as they experienced in their own childhood and from their own father. At the dyad level, this role expectation sometimes resulted in child-rearing conflicts, negative coping strategies, and chronic relationship strain. Findings in this study suggested professional counselling interventions that support positive emotional coping strategies in these dyads effectively reduced

conflict and widened the normative view of these fathers on possibilities to interact with their child. This behavioural shift in the fathers was in turn augmented by strong positive emotional support from their partner spouse. The overall dyadic outcome was a strong sense of “we-ness” and, in two instances, transformative change for fathers. Traditional fathers re-aligned to a new role identity, one Transitional-Traditional and one Transitional-Superdad, over a number of years after their child’s diagnosis and treatment. These fathers were able to move past ideologies and preconceptions of boyhood/fatherhood and accept activities that met the functional abilities of their child. The family-centred model of care provided by the Retinoblastoma Team provided that essential multidisciplinary and professional support throughout the treatment period. Community health professionals also provided these supports to the fathers and their partners.

Hypothesis 3: Fathers experience more social isolation because they lack, or do not avail themselves of, available specific and general social supports.

This hypothesis was true for Traditional, Transitional-Traditional, and Transitional-Traditional fathers. To begin, fathers in all four role definitions identified their partner as the primary provider of social and emotional support during their child’s Rb treatment. Each was highly dependent on their partner for knowledge translation related to their child’s condition. This attenuated fathers’ stress because they better understood the risks and benefits and could actively participate in decision-making with the medical team as required. Fathers in this study indicated they usually relied on informal social support from a small network of peers in the workplace and community, as well as key family members. However, Traditional fathers segmented their peer activities, maintaining regular public-sphere social activities for coping but avoiding or minimizing talking about Rb or their child’s status, even when peers offered informal social support. Transitional-Traditional and Transitional-Superdads preferred to minimize or withdraw from contact with these key peer social supports during early and active Rb treatment periods. These two father types were most heavily reliant on their partner for all informal

social and informational support. Some did, however, actively engage with the Retinoblastoma Team Social Worker for supportive coping during critical periods. Superdads, again uniquely, maintained both familial and outside-the-home peer relationships for social support. Co-worker support was minimized for the periods of time Superdads assumed a full-time caregiving role in the home. Traditional fathers in this study did not access employer or public benefits for personal or spousal support and preferred not to accept instrumental support from community sources. In the same vein, a majority of Transitional-Traditional and Transitional-Superdads minimized the use of work benefits to access social support. Uniquely, Superdads used public-sphere work benefits for professional social supports to attenuate personal and dyadic stressors that negatively impacted their private sphere. Chapter 3 showed cumulative stress and stress proliferation was a reality for many Rb families when their child received chemotherapy treatment. Medically-imposed social isolation from chemotherapy challenged fathers and their partners with loss of personal control. Traditional, Transitional-Traditional, and Transitional-Superdads remained focused on public-sphere work activities during this period, yet reported high levels of treatment-induced social stress. Their partners assumed a primary caregiving role during this time but reported lower levels of social stress from isolation. Superdads in the same situation shared heightened caregiving equally with their partner but had lower levels of social stress like mothers in the same caregiving situation. Actively participating in their child's heightened caregiving needs in this critical period seemed to attenuate social stress for fathers and mothers. As noted earlier, this is suggestive that a 'caregiving as coping' advantage might be considered as an effective parent and dyadic coping strategy in certain contexts.

Hypothesis 4: Rb diagnosis and treatment disrupts the marital dyad at multiple levels, introducing role and relationship changes that are experienced as stressors.

This hypothesis is true, conditioned by disease presentation, the fatherhood role, and regular dyadic coping strategies. Study findings indicated bilateral presentations commonly introduced complexity and uncertainty that in turn heightened individual and

dyadic stress. Caregiving demands were often the first emergent stressor and especially elevated role strain for partners of Traditional and Transitional-Traditional fathers. The ongoing absence of these fathers from attending regular treatment appointments with their partner and child tended to exacerbate partner and dyadic stress, especially when treatment expectations were disrupted. Mothers were often on their own at these moments, sometimes to make critical decisions with the clinician. Negative dyadic coping strategies and chronic relationship strain were a common short-term outcome in many of these situations. Unresolved, this dynamic could devolve into a blame game for couples having a child treated for heritable Rb. Brief face-to-face information meetings with a treating clinician along with Social Work intervention were key elements to attenuate individual and dyadic stress in the short and longer term. Transitional-Superdad dyads fared much better because these fathers made every effort to attend all medical and treatment appointments with their partner and child. The mutual support derived from this symmetrical and positive coping meant these dyads coped better in a crisis and fared well over the longer term. In such a crisis, Superdads quickly moved their focus to the private sphere and shared the entire caregiving role with their spouse. As a result, these dyads maintained existing positive dyadic coping and low-conflict relationships.

Some bilateral parents described their child's Rb as an exceptional experience. This is probably true on two levels – dealing with the disease was an exceptionally challenging coping experience for these dyads and exceptional in the enhanced or intensified relationship the couple experienced throughout the critical period. These dyads were challenged to make sudden, difficult, and sometimes life-changing medical decisions. In general, they experienced a heightened sense of “we-ness” along with mutual trust, tolerance, emotional support, and commitment in the crisis. This heightened or intensified dyadic relationship was transitory in dyads with a Transitional-Traditional father and negative dyadic coping strategies. Transitional-Traditional dyads exhibiting emotion-focused common dyadic coping in the critical period – often supported by external sources like a Social Worker – were likely to maintain that new state of dyadic togetherness. This feeling of “we-ness” was a normative experience in Superdad dyads.

The social and emotional dynamic of sharing and trust permitted ongoing, supportive, reciprocal dyadic engagement across situations and time. These Superdad dyads were unique from the others because they exhibited the core competency for positive systemic-transactional dyadic coping – each spouse was attuned to the other’s ‘stress signals’ and were able to offer reciprocal or situational coping support as needed.

Hypothesis 5: Providing easily understood information on their child’s condition can mediate the experience of stress for both the individual parent and the dyad.

This hypothesis is known to be true from regular and observed clinical practice. The challenge and focus of this research was to uncover insights that inform clinical best practices around psychosocial support for Rb parents. Understanding complex medical information specific to their child, including future outcomes and limitations, was critical to the psychosocial well-being of these Rb parents and families. Fully informed consent to treat is clinically important. Guiding parents towards a clear longer-term understanding of outcomes from treatment is equally important. An optimal solution is staging information for parents, providing multiple sources with consistent messages as their child moves through treatment and follow-up. Time and again parents indicated a strong preference for face-to-face time with the treating clinician. This contact was the primary and trusted source of emotional and informational support for Rb parents. They often perceived limited physician contact time as an outcome of the clinician’s premium role in health care. However, individual clinician preferences also mattered here. Some went beyond factual clinical status updates after each treatment, listening to parents concerns and providing contextual information that supported positive parent and dyad emotional coping in the current period. The real value-added support for parents went beyond just those extra minutes. Effective clinician support involved both acknowledging parents immediate distress and contextualizing stable longer-term outcomes parents might expect as treatment progressed.

Clinical language and complex treatment outcomes were often difficult to understand even with face-to-face clinician meetings and follow-up with other clinical team members. Parents wanted easy access to clear language information sources that reiterated the trusted information from the clinical team. Pamphlets and brochures were a preferred resource, seen as easy to access and understand. These information resources were not readily available to Rb parents at the Hospital for Sick Children, so most used the internet as a primary information resource while acknowledging the challenges of assessing relevance and trustworthiness of content. The family-centred model of care was effective at bridging some of the gap between clinician availability and parent need. A multidisciplinary approach provided both emotional and informational support expertise to parents as needed in the moment. Relatively quick access to clinical and outcome information effectively attenuated individual and dyadic stressors. An ongoing gap for parents, especially mothers, was the desire to network with parents having prior Rb experience. Clinical information aside, mothers wanted to speak to other mothers about life context, to assess the normalcy of their feelings and experiences around their child's treatment. An informal peer network was established to provide matching of Rb families with experienced families when families expressed interest in this type of social support. Geography was often a barrier for matching and some parents did not prefer that level of contact. Social media platforms like Facebook are emerging as the preferred channel for younger mothers to connect with peers for information and social supports.

Implications for clinical practice

Like the child undergoing Rb treatment, parents and families have unique and varied need for psychosocial support that may vary with time and unique treatment challenges. What strongly emerges in this study is keeping fathers engaged and attending regular appointments with their partner and child attenuates a great deal of personal and dyadic stress. This provided dyads with regular connection to the Retinoblastoma Team, especially the Social worker, and opportunity to enhance wrap-around support services with situational interventions to augment positive coping strategies. Findings show Transitional-Superdads and Superdads strongly exhibited this ongoing engagement.

Traditional and Transitional-Traditional fathers quickly disengaged from regular attendance once a treatment schedule was established. These individuals and dyads often experienced higher levels stress related to treatment uncertainty and caregiving. Time and again, parents indicated the Rb clinicians were the primary ‘source of truth’ for emotional and informational support in the treatment setting. That fact seems to offer the best leverage to engage fathers. At intake/diagnosis, consistent messaging from attending clinicians to fathers on the importance of, and reasons for, keeping regular attendance at treatment sessions offers the best chance of compliance. A behavioural nudge strategy could be implemented with a follow-up information/update call or email to a father not attending a treatment session. Best if this came from the attending clinician or delegate (i.e., Fellow) with another Rb Team member as backup. This approach would have to be conditional – often one parent from families living in other provinces travels to Toronto so encouraging the other to attend too would likely impose greater stress on the family. In these cases, a follow-up call from the clinician or alternate to the absent parent after each session would provide the best opportunity to support the dyad and family. This strategy places more up-front demands on already-busy clinicians and follow-up demands on the team. However, a family-centred model of care should adapt to the needs of families and not just maintain regular clinical practice routines. Implementation may require reassessing hospital or clinical policies.

Parents identified face-to-face meetings with clinicians as the preferred channel for disease and treatment information. Often that information, along with chemotherapy and genetic information, clearly overwhelmed parents at first presentation. Understanding this information was especially important for parents’ ability cope and an essential requirement for informed consent to treat. The information-understanding gap created stress for many parents. Regular clinician practice was to refer parents to Rb Team members for follow-up after the information session. Parents found they often had questions outside of regular clinic hours. A preferred source of backup information was clear language topical summary pamphlets and brochures that explained and augmented the detailed information presented in these clinical meetings. Written materials were not

available so parents default information resource for Rb questions was the internet. They regularly accessed a variety of online Rb information while acknowledging there was no way to assess trustworthiness or accuracy of information. It seems judicious to have a single source or truth for Rb treatment in Canada, a website with curated expert content written in clear language for parents. If that is not practical, then moderate the known Rb information websites used by parents and provide a 'curated' and reviewed list of sites containing the most accurate information available on the internet. Multiple channels of information can be embedded in either approach, supporting on-demand information access to Rb mothers who fill a primary knowledge translation role in the dyad and family.

Informal peer social and informational support was an essential need expressed by Rb mothers but not fathers. Mothers speaking to mothers was an essential channel for information and emotional support, especially in the early treatment period. Ad hoc connections were sometimes initiated in the waiting room as children received treatment. An informal peer family-matching program showed some success since implementation. Positive impact was largely limited by geography. Sometimes well-matched families did not live near each other and that desired face-to-face time was limited by both distance and life demands. Findings in this study indicate social media – Facebook – is emerging as the go-to platform for informal peer social and information support. Several participants maintained Facebook websites to describe their own Rb experiences, provide supportive feedback to visitors, and disseminate information on trusted/untrusted information sources. It seems judicious to establish a Facebook page with moderated content to ensure accuracy and trustworthiness of information. A second option is to moderate the know Rb sites and offer feedback, clarification, and alternate resources to participants. Facebook was identified by younger mothers participating in this study. Social media channels are to be of increasing importance as information sources for millennial parents who are coming of age to bear a child with Rb.

Engaging Rb parents through community events, especially an annual research symposium for Rb families has been shown to be an effective engagement activity that includes many fathers. Sharing research from trusted practitioners is an important draw to the event and desired information for parents. The limitation and criticism for organizers was the event was always held in Toronto and Rb families live all over the country. Essentially, ongoing funding is required to host these annual symposiums and either provide live-streaming of the event to Rb families across the country or provide instrumental support for families to attend. The family research symposium would benefit greatly from the implementation of a patient-oriented research program. Having Rb parents/survivors present relevant research findings to other parents would bring an entirely new dimension of engagement to the national Rb community.

Limitations and future research

The opening paragraphs of this chapter discussed possible future sociological research related to moving from descriptive typologies to an explanatory developmental model of fatherhood. Enhanced data collection for fathers is required to link discourse to structural factors in an attempt to develop a robust theoretical model. The discussion thus far has identified a limited number of father-level factors that may offer partial explanation for differences between those with fixed and flexible role adherence over time. Key elements of such a model will likely include elements of family socialization and how masculine identity is referenced in everyday life. In this study, Superdads were unique because they each held a skilled trade certification and lived in multiethnic and multigenerational families co-located in Canada and Western European countries. The two Traditional fathers who also permanently shifted role identity to a Transitional-Traditional or Transitional-Superdad category also held skilled trade certification and had families of origin in Western European countries. The difference was that they did not live that multicultural-multiethnic family experience of Superdads. This researcher speculates that the artisanal training embedded in skilled trades provides these fathers with objective or product measures related to masculine identity. If these fathers also frame caregiving with objective production outcomes, they may be able to enact contested masculine roles

without dissonance. Other fathers in this study were ‘knowledge workers’ in a variety of professional and managerial roles in organizations. For these individuals, masculine role identities may be subjectively governed by organizational performance evaluation processes. The lack of objective production measures limits these father to a recursive process-defined role limited to the organization. In this light, the masculine identity/role is relatively fixed over time. The differential role outcomes of a subjective-objective split in identity measures may hold research value in the masculinities field.

Moving to practical limitations of this study, the epidemiology of Rb does not vary by ethnicity so the lack of representativeness or diversity in this study can be attributed to sampling and recruitment constraints. This was a single-researcher project located at one tertiary care hospital and limited to English-speaking participants. Families receiving care at secondary treatment centres were not excluded but the focus on face-to-face interviews limited the researcher’s ability to engage them in the research. In addition, this study occurred in one of Canada’s largest and most diverse cities yet there were no participants from the global cultural community living in the region. Future research should have representative sampling and the capacity to conduct effective multi-language outreach and research with these diverse ethnic communities. This would permit a comprehensive examination of multicultural communication practices and outcomes related to the family-centred model of care. Expanded understanding of multiple and differing cultural needs and engagement strategies would enhance the ability of the clinical care team to deliver effective wrap-around social and information supports to optimize parent, child, and family outcomes. Second, and related directly to this study, is the ability to recruit more fathers for a deeper understanding of their role identity and coping experiences with critical life events like childhood cancer. Findings here show fathers who stay actively engaged in their child’s treatment process have improved personal coping outcomes over fathers who return quickly to a work focus. Higher levels of positive symmetrical dyadic coping co-occurs with fathers continued engagement in treatment and caregiving. It’s important to understand what treatment teams can do to keep fathers engaged for optimal

individual and dyadic coping outcomes. This should include an exploration of the ‘caregiving as coping’ advantage for other father types.

From a different perspective, how might the emerging context of precarious and contract employment impact the psychosocial and economic well-being of newly affected families in younger cohorts? Findings here indicate families had a range of employer benefits available, whether they used them or not. What if parents had no work benefits or were not routinely supported by employers as was the norm in this sample? How might these lifestyle changes impact delivery of psychosocial care to affected families? Extending the sociological inquiry, we could explore class as a mediator by asking: How would Superdads be impeded from their preferred way of being with their family if they were employed without benefits?

A bigger question for fatherhood research and clinical psychosocial support might be – how universal are these role categories? This research adapted and expanded Cooper’s original 3 fatherhood role identity categories that emerged from work with technology professionals in Silicon Valley. Both studies were located in a North American, post-industrial cultural and economic context. How effectively do fatherhood roles from other cultures map to this categorical model? That knowledge may influence development of specific psychosocial support strategies for newcomer families receiving medical care. To what degree, if any, is the masculine/fatherhood role identity shaped by a combination of education and engagement in organizational work practices? Do subjective performance measures common to organizational cultures provide recursive feedback maintaining traditional masculine roles attached to the public sphere? If so, how can government policies and organizational practices be adjusted to permit flexible fatherhood activities needed to support the well-being of families dealing with a medical crisis?

A key goal of this research was to explore effective parental coping and social support practices and provide insights towards best clinical practices for psychosocial oncology support to parents. Past and existing research was largely survey-based and targeting

mothers in acute care settings. Interviewing fathers was a step towards addressing an existing knowledge gap and providing contextual experiential feedback on supports received in a family-centred model of care. To a degree, mixed methods research designs can substantially inform complex research and add to the body of academic literature in psychosocial oncology. In all these cases, research practice is focused on recruiting representative samples of parents to answer questions developed by experts. What is clearly missing in this domain is a patient-oriented research strategy. This participatory approach engages parents as partners in every phase of the research. No longer just passive participants, parents can inform choice of research questions and design from a unique insider location. They can be active partners in data collection and analysis. Perhaps their greatest impact can be in reporting and dissemination of research findings, adding a unique end-user or consumer perspective to the story. As this dissertation work closes the Retinoblastoma Team embarks on a new national strategy for patient-oriented research. This work will inform that strategy, a good practical end-use for such a large number of words.

Final words

Expected or not, Rb diagnosis introduced an array of sudden and unexpected life course transitions and challenges for families. For some, the impact had intergenerational consequences and concerns. Change was dynamic and often contested for many fathers participating in this research study. This was especially true for those in Traditional, Transitional-Traditional, and Transitional-Superdad categories. Findings support a suggestion by Marks & Palkovitz (2004) that movement or development through diverse styles of fathering may be more the rule than the exception. However, identity change for fathers is conditional and depends on a variety of personal and social support factors.

Parents, individually and as dyads, exhibited resilience in the face of sometimes enormous uncertainty and challenge. Part of that strength comes from readily accessing social and material support from peers, extended families, communities, and the clinical treatment team. Many acknowledged they were personally stronger as a result of this

experience, as was their marriage and their family. The family-centred model of care provides an effective framework to provide interdisciplinary and professional social supports for complex medical cases like Retinoblastoma. Robust implementation in clinical practice can optimize the unique journey and best outcomes for each Rb child and their family.

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Appendix A: Optimal Staffing and Resources for Rb Treatment Centres

| Characteristic | Secondary Rb Centre ≥ 1 | Tertiary Rb Centre ≥ 3 |
|---|-----------------------------------|----------------------------------|
| Number of new Rb patients / year | | |
| Medical school | Recommended | Mandatory |
| Personnel | | |
| Ophthalmic specialist | Recommended | Mandatory |
| Rb specialist | Recommended | Mandatory |
| Back-up ophthalmologist | | Mandatory |
| Pediatric oncologist | | Mandatory |
| Radiotherapist | | Mandatory |
| Pediatric anaesthetist | Recommended | Mandatory |
| Genetic counsellor/Geneticist | Recommended | Mandatory |
| Radiologist | Recommended | Mandatory |
| Referral to an ocularist | Recommended | Mandatory |
| Pathologist with Rb expertise | Recommended | Mandatory |
| Social worker/psychosocial support | Recommended | Mandatory |
| Child life specialist | | Recommended |
| Clinical coordinator | | Recommended |
| Surgical capacity | | |
| Examination under anaesthesia | Recommended | Mandatory |
| Capacity to perform enucleations | | Mandatory |
| Dedicated Rb operating room time | | Mandatory |
| Focal therapy | | |
| Laser therapy | | Mandatory |
| Cryotherapy | | Mandatory |
| Chemotherapy | | Mandatory |
| Radiotherapy | | Mandatory |
| Imaging | | |
| Imaging expertise | Recommended | Recommended |
| RetCam™ | Recommended | Mandatory |
| B-scan 2D ultrasound | Recommended | Mandatory |
| Ultrasound biomicroscopy (UBM) | | Recommended |
| Fluorescein angiography | | Recommended |
| Radiology | | |
| MRI | Recommended | Mandatory |
| CT | Recommended | Mandatory |
| Genetics | | |
| Ability to send samples for genetic testing | Recommended | Mandatory |
| Ocular screening of proband's family | Recommended | Mandatory |
| Research leadership | | |
| Clinical trials | | Recommended |
| Basic science research | | Recommended |

Recommended | Mandatory

Source: National Retinoblastoma Society (2009). National Retinoblastoma Strategy Canadian Guidelines for Care. *Canadian Journal of Ophthalmology/Journal Canadien d'Ophthalmologie*, 44, Supplement 2, S13, Figure 1.

Appendix B: International Intraocular Retinoblastoma Classification (IIRC)

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|--|
| <p>Group A: Very low risk Small discrete tumours not threatening vision (T1a)</p> <ul style="list-style-type: none"> • All tumours are 3 mm or smaller, confined to the retina • Located at least 3 mm from the foveola and 1.5 mm from the optic nerve • No vitreous or subretinal seeding |
| <p>Group B: Low risk No vitreous or subretinal seeding (T1b)</p> <ul style="list-style-type: none"> • Tumours any size or location not in Group A • No vitreous or subretinal seeding • Subretinal fluid no more than 5 mm from tumour base |
| <p>Group C: Moderate risk (T2) Focal vitreous or subretinal seeding; discrete retinal tumours of any size and location</p> <ul style="list-style-type: none"> • Local, fine, and limited seeding (T3) • Discrete intraretinal tumours of any size and location (T2b) • Up to one quadrant of subretinal fluid (T2a) |
| <p>Group D: High risk Diffuse vitreous or subretinal seeding (T3b)</p> <ul style="list-style-type: none"> • Diffuse intraocular disseminated disease • Extensive or “greasy” vitreous seeding • Subretinal seeding may be plaque-like • More than one quadrant retinal detachment |
| <p>Group E: Very high risk (T4a) Very high risk with one or more of the following:</p> <ul style="list-style-type: none"> • Irreversible neovascular glaucoma • Massive intraocular hemorrhage • Aseptic orbital cellulitis • Tumour anterior to anterior vitreous face • Tumour touching the lens • Diffuse infiltrating Rb • Phthisis or prephthisis |

Adapted from: National Retinoblastoma Society (2009). National Retinoblastoma Strategy Canadian Guidelines for Care. *Canadian Journal of Ophthalmology/Journal Canadien d'Ophthalmologie*, 44, Supplement 2, S23, Table 7.

Appendix C: Retinoblastoma Parent Interview Instrument

Part A: First, I would like to explore events leading up to (CHILD)'s diagnosis.

1. Was there any prior indication of an eye problem?
2. Who first noticed the problem and what did they report?
3. Were you present when (CHILD) was diagnosed with Retinoblastoma?
If 'yes', skip to question 6 If 'no', continue to question 4
4. Where were you at that time?
5. How and when did you find out about the diagnosis?
6. Had you ever heard of Retinoblastoma before that moment? What was your initial reaction?
7. How long did it take to understand the serious nature of Retinoblastoma?

Probe: Were any individuals particularly important in helping you understand Retinoblastoma?

Probe: What other information sources did you use?

8. What are the biggest challenges/concerns/problems that you [deal/dealt] with during (CHILD)'s treatment?

Probe: Travel costs? Care or work arrangements? Understanding and acceptance?

Go to Part B.

Part B: I would like to ask some questions on your background.

9. What is your birth year?

10. Were you born in Canada?

If 'yes', skip to question 14

If 'no', continue to question 11

11. What was your country of birth?

12. When did you immigrate to Canada?

13. What is your current citizenship status?

Probe: Refugee? Immigrant? Landed immigrant? Citizenship?

14. How do you describe or identify your ethnic background?

Continue to Part C.

Part C: Next, I have a few questions about your employment status.

15. Are you currently employed?

If 'yes', skip to question 18

If 'no', continue to question 16

Not in labour market:

16. Are you currently a full-time parent or care provider or student?

17. Are you actively seeking paid work?

Continue to question 38, Part D.

Full-time employment:

18. Is this a regular full-time position?

If 'yes', continue to question 19

If 'no', skip to question 25

19. Describe the type of work are you doing?

Probe: What kind of business or industry is this? What do you make or do at work?

20. How long have you held this position?
21. Do you manage people, money, information or technology?
22. How many hours per week do you usually work, including regular overtime?
23. Do you belong to any work-related professional organizations?
24. Has your employment ever been disrupted by job loss, health, or family issues?

Continue to question 38, Part D.

Part-time employment:

25. Do you work at a part-time job?

Probe: Regular, contract, seasonal, occasional?

26. Briefly describe the type of work are you doing?

Probe: What kind of business or industry is this? What do you make or do at work?

27. Do you manage people, money, information, or technology?
28. How long have you held this position?
29. How many hours per week do you usually work, including regular overtime?
30. Has your employment ever been disrupted by job loss, health, or family issues?
31. Do you belong to any work-related professional organizations?

Continue to question 38, Part D.

Part D: Now, A few questions on work-related benefits.

32. Briefly describe any work-related benefits you receive (or have access to)?
- Probe: paid sick leave; parental leave; flexible hours or flex time; work from home?
33. Have you used these benefits to spend more time with (CHILD) during treatment?
34. Have you used these benefits to spend more time with your spouse/partner?
35. Have you accessed these benefits yourself, for your own well-being?
36. Would you be willing to tell me your total annual household income, before taxes, to the nearest \$10,000.

Continue to part E.

Part E: Next, I would like to ask you some questions on your education.

37. What is the highest grade or year of school that you have already completed?
- Probe for certificates, diplomas, or degrees.
38. Have you done any continuing education or job-related training since leaving school?
- Probe: Was this training required for promotion or continued employment?
39. Some people complete all their schooling and then enter the workforce. Others move between school and work. Describe your pathway.
- Probe: length of time between transitions? Unemployment?

Continue to Part F.

Part F: In the next part, I have some questions on your marital status.

40. Are you currently married or living common law?

If 'yes', continue to question 41

If 'no', skip to question 42

41. How long have you been married [lived common law] (years)?

Go to question 52, Part G.

42. Are you currently separated from your spouse/partner?

If 'yes', continue to question 43

If 'no', skip to question 44

43. How long did you live together (years)?

Go to question 49

44. Are you currently divorced?

If 'yes', continue to question 45

If 'no', skip to question 46

45. How long?

Go to question 49

46. Are you a single (never married) parent?

47. Do you have any contact with (CHILD)'s biological parent?

48. Are you the primary parent?

If 'yes', skip to question 52, Part G

If 'no', continue to question 49

49. How often do you see (CHILD) (during week, weekends, holidays)?

50. Are there any barriers to spending regular time with (CHILD)?

51. How is (CHILD)'s health information generally communicated to you?

Probe: Did you participate in (CHILD)'s Retinoblastoma treatment decisions?

Probe: How and when were you informed of diagnosis and treatment decisions?

Continue to Part G.

Part G: Next, I want to determine your relationship with (CHILD).

52. Are you (CHILD)'s biological mother/father?

If 'yes', continue to question 53

If 'no', skip to question 55

53. Was this a planned pregnancy?

54. How old were you when (CHILD) was born?

Got to Question 61

55. What is your relationship to (CHILD)?

56. How long have you lived with (CHILD)?

57. Do you have any other biological or adopted children?

58. Do they live with you? If not, who do they live with?

59. Do you have regular contact with them?

60. Have there ever been periods of one week or more when you were away from home or (CHILD) was away from home?

Probe: child's illness/treatment, parent work or illness; marital disruption; vacation?

Continue to Part H.

Part H: Now I would like to ask you about your social supports.

61. Did you attend prenatal classes with your spouse/partner? Why or why not?
62. Did you discuss issues or concerns about the pregnancy with your spouse/partner?
63. Thinking of (CHILD)'s birth, were you present when he/she was born? Why or why not?
64. Have you ever attended a meeting, event, or program that related to raising children or being a better parent? Why or why not?
65. Have you ever attended a meeting that was just for [fathers/mothers]? Why or why not?
66. Are there other individuals you can turn to when you are unsure how to deal with a parenting issue?

Probe: What is this person's relationship to you?

67. In what ways does your spouse help you to be a good parent?
68. How often do you talk to your spouse/partner about (CHILD)?
69. How often do you talk to your spouse/partner about your day or things that interest both of you?
70. Has (CHILD)'s Retinoblastoma changed communication with your spouse/partner?
71. Has Retinoblastoma changed getting together with family, friends, or neighbours?

Continue to part I.

Part I: Finally, a few questions on how you see yourself as a parent.

72. What practical, everyday things do you do to be a good parent? Can you give me some examples?
73. What gets in your way of being the kind of parent you'd like to be?
Probe: How do these (people/things) stand in your way?
74. How important is it to you that you are a good provider to (CHILD)?
Probe: Do you ever give up time with (CHILD) to focus on work and earnings?
75. How do you fit work and family time together?
76. How often do you take on the primary caregiving role for (CHILD) while your spouse/partner does other things? How long are these periods of time?
77. Has (CHILD)'s Retinoblastoma changed how much daily care you are required to provide?
78. Has it changed your spouse/partner's active participation in (CHILD)'s daily care?

Thinking about (CHILD) and yourself over the next few years:

79. What are your major concerns for him/her, now and in the next few years?
Probe: current and future health issues, education, sports, social adjustment and peer acceptance, secondary cancer, skill and employment, risk to future offspring?
80. How will you help (CHILD) deal with these challenges?
81. That is all the questions I have today. Is there anything you want to add? Do you have any questions on this research project?

Curriculum Vitae

Name: **Robert Downie**

Post-secondary Education and Degrees

King's University College
London, Ontario, Canada
1982-1986 B.A.

University of California Extension
Los Angeles, CA, USA
1999-2001 Certificate in Business and Technical Writing

Western University
London, Ontario, Canada
2002-2003 B.A Sociology (Hons.)

Western University
London, Ontario, Canada
2003-2005 Completed M.A. coursework, accelerated to PhD

Western University
London, Ontario, Canada
2005-2017 PhD Sociology

Honours and Awards

Province of Ontario Graduate Scholarship
2005-2006, 2007-2008

Social Science and Humanities Research Council (SSHRC)
Masters Fellowship 2004-2005
Doctoral Fellowship 2006-2007

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|--------------------------------|---|
| Work Experience | Manager, Institutional Research Fanshawe College 2014-current |
| | Senior Researcher Fanshawe College 2010-2014 |
| | Analyst, Research and Evaluation Unit Surrey Place Centre, Toronto. 2009-2010 |
| | Graduate Research Assistant Western University 2003-2004 |
| | Research Assistant Acupuncture Institute 1987-1999 |
| Related Work Experience | Instructor, Research Methods Brock University. 2010-2012 |
| | Cultural Competency Research (contract) Canadian Forces Leadership Institute 2009 |
| | Indigenous Disability Research (contract) Aboriginal Affairs and Northern Development Canada 2008 |

Research Associate, APRC(I)

Western University

2005-2007

Graduate Research Associate

Workforce Aging in the New Economy (WANE)

2003-2004

Peer-reviewed publications:

Chiu, H. H., Dimaras, H., Downie, R., & Gallie, B. (2015). Breaking down barriers to communicating complex Retinoblastoma information: can graphics be the solution? *Canadian Journal of Ophthalmology/Journal Canadien d'Ophtalmologie*, 50(3), 230-235.

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Presentation Committee, Primera Cumbre Mundial de Armonización de Medicina Tradicional, Alternativa y Complementaria (2008). Declaración de Lima. *Acta Médica Peruana*, 25(2), 123-124.

McKibbin, L.S. & Downie, R. (1995). The Effects of Low Energy Laser on Soft Tissue in Veterinary Medicine. *Laser Therapy* 7, 42-51.

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Select articles, reports, and working papers:

Downie, R. (2010). Sustainable Cancer Treatment in Kenya. *Juxtaposition Global Health Magazine*, 4(1), 10-12. Toronto, ON: University of Toronto.

White, J., Peters, J., Spence, N., & Downie, R. (2007). *Aboriginal Labour Market Participation in Canada, United States, New Zealand and Australia: Technical Report*

APRC (I) 2007-01. Aboriginal Policy Research Consortium (International): The University of Western Ontario.

White, J., N. Spence, J. Peters, and R. Downie (2007). *Aboriginal Labour Market Participation in Canada, United States, New Zealand and Australia: Assessing the current situation and lessons that can be drawn.* Aboriginal Policy Research Consortium (International): The University of Western Ontario, Working Paper 2007-01, pages 34-60.

McMullin, J. A., Cooke, M., & Downie, R. (2004). *Labour Force Ageing and Skill Shortages in Canada and Ontario.* Research Report W|24 Work Network, Ottawa: Canadian Policy Research Networks.

Downie, R., Dryburgh, H., McMullin, J., & Ranson, G. (2004). *A Profile of Information Technology Employment in Canada.* WANE International Report #1, London: Workforce Ageing in the New Economy, The University of Western Ontario.

Invited presentations:

Regulating and Integrating TACM: The Canadian Experience. Cumbre Mundial de Armonización en Medicina Tradicional, Alternativa y Complementaria (World Summit for Harmonization of Traditional, Alternative and Complementary Medicine). Lima, Perú, November 7-11, 2007.

Conference presentations:

Comparing factors in the KPI Satisfaction and i-graduate surveys. KPI+ Workshop, Ministry of Training Colleges and Universities, Seneca College, Toronto, June 2013.

Modeling College Student Engagement using 2010 KPI Satisfaction Data. KPI+ Workshop, Ministry of Training Colleges and Universities, Humber College, Toronto, June 2011.

Personal and Marital Stressors When a Child is Diagnosed With Retinoblastoma. Canadian Retinoblastoma Society Research Day, Toronto, June 2009.

Monitoring outcomes in child and adolescent psychotherapy. Research Special Interest Group – Ontario Association on Developmental Disabilities Conference, April 9, 2010. Toronto, Ontario.

Personal and Marital Stressors Occurring When a Child is Diagnosed With Eye Cancer. Annual Sociology Graduate Student Conference. University of Western Ontario, London, Ontario. April 30, 2009.

The Impact of Social Structure on Population Health Measures. Global Tensions, Global Possibilities Conference. McMaster University, Hamilton, Ontario, September 21-23, 2007.

Improving Complex Cancer Care Decisions For Children. International Sociological Association RC-06 Health in Families, Healthy Families: Gendered Explorations. University of Toronto, Toronto, Ontario, May 8-11, 2007.

Preliminary Results from the Family Needs Survey for Retinoblastoma. National Retinoblastoma Strategy Meeting, Toronto, November 2006.

Evaluation Issues with Electronic Health Records. Cancer Informatics Department, Princess Margaret Hospital, Toronto, Ontario, May 2005.

External funded research:

Co-investigator (Fanshawe), *Essential Adult Skills Initiative*. Higher Education Quality Council of Ontario. 2015-2016. \$29,200.

Third-party Evaluator, Local Employment Planning Council Pilot. 2015-2016. \$23,400.

Principal Investigator (Fanshawe). *Piloting the Collegiate Learning Assessment and the Community College Learning Assessment*. Higher Education Quality Council of Ontario. 2011-2013. \$41,376.

Co-investigator, *Outcomes of structured psychotherapy intervention for high-functioning adolescents with autism*. Supporting Service Excellence and Continuous Quality Improvement Through Evaluation Grant Initiative, Ministry of Community and Social Services. 2010. \$36,450.

Related committee work:

Heads of Institutional Research, Colleges Ontario, Toronto. 2014-current.

Research Committee, CURA2 – Poverty and Social Inclusion Project, Lawson Research Institute, Western University. 2014-2016.

Team 3 Lead, Survey Tool Development. *Impact evaluation of youth livelihoods development project in southern Iraq*. Statistics Without Borders/Save the Children. 2013.

Research Ethics Board, Surrey Place Centre, Toronto. 2009-2010.

Research Officer, Daisy Fund, Hospital for Sick Children, Toronto. 2007-2009.

National Retinoblastoma Strategy, Hospital for Sick Children, Toronto. 2005-2008.

Peer reviewer:

Canadian Journal of Program Evaluation.

International Journal of Sociology and Social Policy.

The International Indigenous Policy Journal.

McGill Sociological Review.

Professional memberships:

Canadian Evaluation Society.

American Evaluation Society.

Australasian Evaluation Society.

European Evaluation Society.

Canadian Institutional Research and Planning Association.

Association for Institutional Research.