University of Alberta

A randomized controlled trial of storytelling as a communication tool aimed at parents of children presenting to the emergency department with croup

by

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This work is dedicated to the memory of my mother,

Lois Hartling (1933-2007)

To every thing there is a season and a time to every purpose under the heaven.
Ecclesiastes 3:1

Abstract

Background: Stories may be an effective tool to communicate with and influence patients because of their ability to engage the reader.

Objectives: To develop story booklets and evaluate their effectiveness compared to standard information sheets for parents of children attending the emergency department (ED) with a child with croup.

Methods: A systematic process was followed to develop and pilot-test the story booklets. Parents were randomized to receive story booklets or standard information sheets during their ED visit. The primary outcome of change in anxiety during the ED visit was assessed using the State Trait Anxiety Inventory, which was completed upon recruitment and at discharge. Follow-up telephone interviews were conducted at 1 and 3 days post-ED visit to gather information on secondary outcomes: symptoms, expected anxiety for future croup, satisfaction, regret, knowledge, return for medical care, and resource use. Telephone interviews were conducted every other day until symptoms resolved or until day 9. Outcomes were compared using independent-groups t-tests, Mann Whitney tests, or Chi-square tests.

Results: There was no significant difference in the primary outcome of change in parental anxiety between recruitment and ED discharge. The story group (n=129) showed significantly greater decision regret regarding their decision to go to the ED than the comparison group (n=126) ($p<0.001$). The story group reported quicker resolution of symptoms: median days to no symptoms 3 versus 5; the survival distributions were significantly different ($p=0.032$). There were no differences for the remaining outcomes.

Conclusions: This study provides preliminary evidence regarding the use of stories in the ED for an acute, self-limiting condition and contributes to a growing evidence matrix identifying when, where, and for whom storytelling may be most effective. Reasons for lack of significance for the primary and other outcomes

may relate to choice of outcome, timing of outcome assessment, or disconnect between the intervention and needs of the end-user. Further research is needed to corroborate the significant findings and examine their underlying mechanism. An examination of risk of bias in a sample of pediatric trials demonstrates that there is room for improvement in the design, conduct, and reporting of research related to child health and provides direction for future research.

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Chapter 1

Introduction

1.1 Overview of the problem to be addressed

Children's illness and injury cause parental anxiety, even with common and self-limiting conditions and particularly among the younger age groups.[53,66] Major sources of parental anxiety are uncertainty due to a lack of knowledge about the condition and its management.[49,101] The provision of information related to the illness and processes of care has been linked with reduced anxiety and uncertainty as well as greater satisfaction with medical services and more appropriate healthcare utilization.[66,101] The increasing demand for consumer-friendly, reliable health information has prompted extensive research to identify effective methods of communication. Standard written instructions, used in many clinical settings, have been found wanting,[79] while alternative formats (such as video presentations, illustrations, and cartoons) have been found to be more effective.

The effectiveness of storytelling as a communication tool has been supported by evidence from several disciplines including nursing, social science, and psychology.[61] An appeal of storytelling is its ability to present information couched within a personal account that engages the reader and validates their own experiences. Further, stories presented in plain language may be more understandable to a lay audience.[78] This method may be especially appropriate in the pediatric emergency department (ED) where the busy setting can be anxiety-inducing for parents and their children, parental anxiety can be high related to their child's condition, and time spent with health care professionals can be brief. The primary aim of this study was to investigate storytelling as a tool to engage parents in communicating research and health information in order to affect parental anxiety and other outcomes.

The terms "narrative", "story", and "storytelling" have been used variably, and at times interchangeably, in the literature. Hinyard specified that narrative has "an identifiable beginning, middle, and end that provides information about scene, characters, and conflict; raises unanswered questions or unresolved conflict; and provides resolution."[73] A story is considered the "retelling of an experience or a

fictional account of an experience."[73] Both genres may contain messages with an intent "to teach, or convey something to the listener."[98,138] One distinction that has been made between narrative and story is that narrative has plot in its structure (explains why something happened) whereas a story is a simpler recounting of events (explains what happened).[51,60] In general, the distinction between narrative and story appears unclear with substantial overlap between the two. In a recent relevant publication the two terms were used interchangeably and given the same definition.[64] Storytelling is "a distinct and unique method for making stories available to others."[111] In this study we investigate the use of storytelling, or making real-life stories available, to communicate health information to parents (or the child's primary caregiver). We focus on the terms "story" and "storytelling" when referring to this study and our hypotheses, but use the term "narrative" when referring to reports wherein the term was used.

1.2 Rationale for the trial

Almost a quarter of Canadian children seek emergency care in any given year.[153] Attending the ED is an anxiety-provoking experience for children and their parents.[66] One of the major sources of parental anxiety is "uncertainty about what will happen at the hospital and unanswered medical questions."[49,101] Flury et al. showed that severe anxiety among parents accompanying their children to the ED was significantly associated with lack of knowledge.[50] This anxiety can be heightened by prolonged waiting, unfamiliar surroundings, technical equipment, interactions with medical professionals, and lack of control.[56,129] Parents may also be anxious due to their child's discomfort, the basic procedures performed on their child, and the prospect that their child may experience ongoing harm from the presenting illness.[129] Addressing parental anxiety can impact more than the parent's experience; parental anxiety is thought to have detrimental effects on the child.[6,49,101] For instance, there is some evidence suggesting that parents witnessing painful procedures in their child have elevated heart rate, blood pressure, and anxiety and that in turn parental anxiety can contribute to the child's anxiety and perceived pain.[108,135,145,149]

The provision of information about an illness and its management is associated with reduced anxiety, enhanced knowledge, and increased satisfaction with care. Providing timely and useful information to parents can assist in managing their anxiety.[50] Informing and preparing parents and children of what to expect is also linked to parent satisfaction, compliance, and cooperation during and after the ED visit.[49] Trout et al.'s review of patient satisfaction in the ED showed that satisfaction was strongly associated with positive provider-patient communication, efforts to enhance patients' understanding of care and processes of care, information provided to the patient, and meaningful communications between the patient and staff.[158] The importance of communication is also reinforced in a prospective study that evaluated the provision of information to adult patients upon their arrival to the ED. Compared to a control group that received no information, the patients in the experimental group were significantly more satisfied overall and rated specific aspects of care significantly higher including physicians' care and concern, ability of the staff to decrease patient anxiety, physicians' explanation of illness and treatment, and the information provided.[100]

Standard written instructions are not as effective as more innovative methods of presenting information. The anxiety that stems from uncertainty and lack of knowledge has motivated the development and provision of educational materials for pediatric patients and their families. The format for delivering the information has been the subject of previous research. Standard written instructions, used in many EDs, have been found to be ineffective.[79] For instance, in a study of the use of routine computer-generated discharge instructions, the majority of the parents who had attended a pediatric ED did not recall receiving any information 1 to 2 weeks after the visit.[134] An important barrier to effective communication is the use of medical terminology and quantitative information, which is often not well understood by patients and their families.[33,56,115,163] According to the Institute of Medicine in the US, almost half of American adults have limited health literacy

which results in less preventive health care and more frequent use of expensive health services.[78]

Effective communication involves transforming the information into messages that can be easily understood and readily accepted by the intended audience.[144] A randomized controlled trial (RCT) comparing wound care instructions with and without cartoons found that the group receiving the instructions with cartoons was more likely to have read the instructions, to answer all wound care questions correctly, and were more compliant with daily wound care.[42] Another RCT demonstrated that instructions accompanied by illustrations for patients discharged from the ED with lacerations enhanced patient comprehension compared to the same instructions without illustrations.[8] This evidence suggests that, to be most effective, the information must be presented in a format that is engaging and understandable.

Stories have the potential for reducing anxiety and enhancing knowledge and satisfaction with care because they are "believable, rememberable, and entertaining."[125] Storytelling is one of the oldest forms of communication and is "an intrinsic part of most cultures."[110] In the past century of Western medicine, however, its use has been overshadowed by more objective approaches including reliance on modern technology.[30] The result has been concern that "doctors do not listen to their patients" and that doctors "seem unmoved by what their patients experience."[31] Recently, there has been resurgence in the use of storytelling in medicine in diagnostics,[31,148] therapeutics,[32,142,150] and the education of patients, students, and practitioners.[12,30,91,106,131,164] This movement is attempting to provide a more holistic and intuitive approach to patient care. Verghese described two parts to an illness, "a physical deficit and a spiritual violation."[160] Factual information can address treatment and expectations for the physical component, but "does not address the social, emotional, and motivational influences of illness."[160] Stories present an opportunity to address simultaneously both the physical and spiritual aspects of an illness.

Stories are an integral part of learning [130] and play a key and powerful role in our education and development from an early age. Stories are effective because they "appear to be processed in an automatic, relatively effortless way and are associated with efficient processes of memory and retrieval."[147] Anecdotal evidence suggests that stories provide a means for realization of "sameness" and that feelings are acknowledged and validated.[154] Stories generate more impact than simple statements of fact because of the situation-specific details and human experience related through the recounting of events;[137] further, information and details are recalled longer if they have an emotional impact.[1] Underlying the hypothesis for this study is that stories would have a similar effect for parents of sick children by providing context to deliver information related to their child's condition and management.

Summary: The hypothesis for this trial was motivated by the four factors described above: (1) attending the ED with a child is anxiety-provoking for the parent; (2) provision of information can reduce anxiety and increase satisfaction with care; (3) to be most effective the information must be delivered in a form that is understandable and engaging; and (4), storytelling may be a powerful tool to communicate with parents and their children. Our goal was to investigate storytelling as a communication tool for health information while attending to the personal experience of the parent and pediatric patient.[26]

Clinical context for the trial: Croup was chosen as the condition with which to examine the hypothesis because of the frequency of its presentation to the ED,[22,43,70,116] the anxiety that it causes for parents,[41] and the large body of evidence that supports the therapeutic management of the disease.[2,17,21,37,48,55,82,86,94,105,127,155,156,165] Croup is a common respiratory tract illness most often affecting children between 6 months and 3 years of age.[43] Croup causes much anxiety for parents, largely due to the nature of the cough (barking), difficulty breathing (hoarseness, inspiratory stridor), timing of onset which often rouses children (and parents) from their sleep late at night, and their lack of knowledge regarding the condition.[41] In a survey of parents accompanying

their children to the ED with mild croup, a large proportion reported being "very concerned" with: the child's respiratory effort (72%), unusual sound of breathing (69%), potential lack of oxygen (59%), unusual sound of cough (53%), and painful uncomfortable cough (55%).[41] Further, parents expressed intense concern over their lack of knowledge about croup (45%). A subsequent study confirmed these findings and showed that over 60% of parents were afraid that the child might stop breathing while 20% were extremely concerned that their child might die (unpublished data; personal communication, DW Johnson, Professor, Department of Pediatrics, University of Calgary). Further, 50% of parents expressed concern over their increasing tension and frustration as a reaction to the situation. From a practical perspective, parents were very or extremely concerned that their child might be hospitalized (52%), that the illness might recur (60%), and their lack of knowledge regarding the illness (40%).

1.3 Literature review

Two medical research librarians (Lisa Tjosvold and Carol Friesen, Alberta Research Centre for Health Evidence, Department of Pediatrics, University of Alberta) with experience in search methods for systematic reviews conducted a comprehensive search of the literature. Ten electronic databases were initially searched in 2006; the search strategy for each database is detailed in Appendix A. The search yielded 117 citations. Upon review of the abstracts, 75 studies were found to not be relevant, while 42 studies were reviewed in detail for potential relevance to the research question. An updated search restricted to PubMed and Dissertation Abstracts was conducted in June 2009 to identify recent publications. The updated search yielded 74 citations of which 33 were reviewed for potential relevance.

One of the findings from the literature review is that both fictional and non-fictional stories are used in medicine. The focus of the present research is on stories that are based on the real-life experiences of patients and their parents as opposed to stories that are fabricated for specific purposes.[e.g.,19,36,114] The review

also revealed that stories are being used in many different forms, as well as for a wide variety of conditions and situations. For example, storytelling has been developed: in the context of theater as a method to share knowledge regarding cancer-related issues and influence behavior among Alaska Natives;[40] as a tool for risk reduction among drug-using women in inner-city American communities;[136] as an educational and supportive resource for patients living with arthritis;[154] as a resource to support stroke patients and people close to them;[162] as a web-based tool to aid in decision-making related to screening for colorectal cancer;[80] within patient decision aids;[89,159] and, in the context of persuasive messages to encourage people to carry signed and witnessed organ donor cards.[96] Storytelling also is commonly employed as a tool for educating vulnerable target populations about HIV/STD prevention.[14] Recently stories have been investigated in the context of questionnaires to measure well-being and cultural adherence.[63]

Stories have been used in an assortment of other clinical areas including: child psychotherapy,[15] children with critical illness,[52] older adults with chronic illness,[23] diabetes education,[62] heart disease,[109,117] mental health,[112] and various forms of cancer.[33,39,47,99,103,104] Other modalities have also been used in combination with the storytelling technique such as drama, song, conversation,[107] and role modeling.[77] Petraglia has distilled the forms in which narrative is being used into two broad categories: narrative therapy versus narrative intervention.[132] Narrative therapy refers to a therapeutic technique where the patient creates and shares their own narrative, primarily to enhance their ability to cope with an illness; a narrative intervention is created by an independent party and provided to the patient to effect change for a variety of outcomes such as attitude, knowledge, and behaviour.

There are no systematic reviews in this area. Moreover, few studies have evaluated narrative/stories in randomized trials and among the trials that exist, there is variation in the purpose of the stories and target populations. Most of the studies have been conducted in the context of health promotion and disease

prevention for a variety of medical conditions. The following is a summary of the trials identified in the area of storytelling.

Only one trial involved a pediatric, clinical population and addressed management of parental anxiety. Melnyk et al. developed a 3-phase educational-behavioural intervention for children admitted to a pediatric intensive care unit and their mothers which included reading and discussing a story about a young child who successfully copes with a stressful hospitalization.[119] The intervention was tested in a randomized trial involving "174 mothers and their 2- to 7-year-old children who were unexpectedly hospitalized in the pediatric intensive care units." [119] While there were no differences in parental anxiety during hospitalization, the intervention group showed reduced anxiety (effect size of 0.32 at 1 month post-discharge). The intervention also reduced depression, and symptoms of post-traumatic stress disorder following the hospitalization. The largest effect sizes were seen in the follow-up period after hospitalization, potentially due to differential loss-to-follow across groups (overall 58.2% attrition rate by 1 year post-hospitalization with control group showing more missing data over time). One of the study's limitations was that it was not possible to isolate the impact of the storytelling component among the other facets of the complex intervention.

Noell et al. developed and tested interactive videodisc programs to "teach decision-making skills and socially appropriate responses" in order to reduce HIV/STD risk behaviours among adolescents.[126] The videodiscs followed a storyline and provided education around risk behaviours (e.g., safe sex, condom use, etc). The authors tested the programs in a cluster randomized trial involving 47 classrooms (827 students) in terms of beliefs, intentions and attitudes, and self-efficacy. Outcomes were assessed immediately and 30 days after receiving the intervention. Classrooms were randomized to the intervention or wait-list control; the intervention was viewed during a single class session. The results showed significant differences on all variables at either the immediate post-test or 1 month follow-up, suggesting the intervention was "effective in changing attitudes, intentions, and self-efficacy related to sexual behaviours." [126] The limitation of

this study is that there was no information on actual behaviours or ultimately changes in rates of HIV/STD infections. Feedback from the participants highlighted the importance of interactivity of the videodisc program and matching the materials to student ethnicity.

Slater et al. examined the effectiveness of testimonial, conversational, and didactic formats for providing nutritional information in terms of believability, clarity, perceived usefulness, and self-efficacy.[147] It was hypothesized that these factors are pre-requisites for effecting behavior change. The authors employed a Greco-Latin square experimental within-subjects design where all 31 participants received each intervention format in random order. The researchers found that the conversational format was significantly more believable, but no differences were observed for clarity, perceived usefulness, or self-efficacy. The authors commented that "the ability to detect differences was limited by the generally positive ratings of the messages, which reduced variability." [147] The authors concluded that the main reason the narratives did not perform better was that they lacked emotional engagement, or the ability of the reader to identify with the people in the story (i.e., the source of the message).

Larkey et al. conducted a pilot quasi-randomized (alternate allocation) trial to compare storytelling versus a numeric risk tool to convey health promotion information about colorectal cancer prevention to the Latino population.[103] Trained health educators delivered the interventions over a 30-45 minute session after which post-intervention surveys were administered. The outcomes of interest were intention to change behaviors, including increased consumption of vegetables, increased daily physical activity, screening for colorectal cancer, and encouraging others to screen. Significant results favouring the storytelling intervention were found for intention to increase vegetable consumption and physical activity. There were no significant differences in measures of fear of colorectal cancer, perceptions of risk, intent to screen or to encourage others to screen. The authors suggested that for some comparisons the small number of

respondents (n=64) made comparisons not meaningful. The study did not collect information on actual behaviour change or long-term outcomes.

In 2009, Larkey published results of another pilot randomized trial similar to the first reported, although the second study was restricted to women (n=78).[104] The storytelling group was significantly more likely to intend to screen and encourage others to screen. No significant differences were found for intent to increase physical activity, intent to increase vegetable consumption, or perceived risk or fear of colorectal cancer. The study had the same limitations as the first in terms of no measures of actual behaviour change and no long-term follow-up.

Mazor et al. conducted a randomized trial to compare three methods of communicating information related to anticoagulant therapy and monitoring.[115] The interventions were videos involving a physician-patient encounter with three different contents (narrative, i.e., patient anecdotes; statistical evidence; or both). These were compared to a "usual care" group. The outcomes of interest were patients' knowledge, beliefs, and behaviours. In the end 317 patients of 592 who agreed to participate returned both baseline and follow-up questionnaires. All groups showed significant improvements in knowledge and certain beliefs compared to the control group. When comparing the narrative and statistical videos, the narrative group showed significantly stronger belief that lab testing is important; knowledge was greater in the narrative group only when controlling for baseline knowledge score. No differences were noted between narrative video and video with both narrative and statistical evidence. While there was some evidence to support the narrative format, results were not consistent across all outcomes. The authors concluded that "findings are promising, but clearly far from definitive. Clearly, there is a need for future empirical work that systematically investigates the factors that influence whether or under what conditions narrative evidence has an impact, what that impact is, and what the relevant interactions are."

McDonald et al. compared factual versus storytelling formats to teach women "how to recognize and respond to symptoms of" a myocardial infarction.[117] The study also evaluated cognitive restructuring the social norm of "caring for others" to "caring for self" and evaluated these two variables (format and cognitive structure) in a factorial pretest-posttest randomized design. A total of 113/120 women completed the study. No significant differences were found across groups for learning MI symptoms or intention of calling 911 if MI symptoms occurred, although all participants had high baseline intention of calling 911. The authors cited as a limitation the fact that participants did not necessarily take the time to read the pamphlet. Further, outcomes were assessed immediately after reading the pamphlet; hence, there were no data on long-term outcomes or whether the intervention had an impact on actual behaviour.

Overall, the literature illustrates that storytelling is being sought as a tool to communicate with and influence patients or at-risk populations. It is not possible to make general conclusions around the effectiveness of stories due to the heterogeneity between studies in populations (including settings and clinical conditions), interventions, comparisons, and outcomes. There are several general limitations with the individual studies including lack of data on long-term outcomes and end-point outcomes. The studies vary substantially in size from 31 to 847 participants; however, only one study provided sample size calculations.

Most of the previous trials were at high [103,104,115,119,147] or unclear [117,126] risk of bias in their estimates of effect.[71] Risk of bias was unclear for sequence generation in five studies,[104,115,119,126,147] high in one study,[103] and low in one study.[117] Risk of bias for allocation concealment, was unclear in four studies,[104,115,126,147] high in two studies,[103,119] and low in one study.[117] Four studies were at high risk of bias due to lack of blinding,[103,104,119,147] two studies were unclear,[115,126] and one study blinded the data collectors and data analysts.[117] Two studies adequately addressed incomplete outcome data,[104,117] while this was unclear in three studies;[103,126,147] two studies had either substantial loss to follow-up [115] or differential loss to follow-up over time.[119] Risk of bias due to selective

outcome reporting was low in four studies [103,104,115,119] and unclear in three studies.[117,126,147] Finally, most studies were free of "other sources of bias",[103,104,115,117,119,147] while one study was unclear.[126]

The paucity of rigorous clinical research studies underscores the need for additional evidence to confirm or refute the value of stories or storytelling as a communication tool within the healthcare setting, and specifically in the context of pediatric care.

1.4 Research Question, Objectives and Hypothesis

The principal research question is: Can we affect parental outcomes and resource use through stories that integrate research and health information with personal experience?

The hypothesis was that stories, delivered through printed and illustrated story booklets, versus standard information sheets distributed in the ED, would produce different results in terms of parental anxiety, knowledge, satisfaction, and decisional regret; healthcare utilization patterns; and resource utilization.

The primary objective was to evaluate the effectiveness of story booklets compared to standard information sheets in terms of parental outcomes, child symptoms, and resource use.

The second objective was to examine the evidence for storytelling from randomized controlled trials in the context of other pediatric trials and risk of bias.

Chapter 2

Development of Story Booklets

2.1 Background

While there is a growing body of literature discussing the use of stories and storytelling as a communication tool in healthcare or health promotion, there are few accounts describing the development of the interventions including detailed testing among the end-user group. Of the previous randomized trials in this area presented in Chapter 1, only one described extensive formative research, including involvement of the target population in the development and testing of the intervention.[126] One study stated that formative research was not possible due to time and resource constraints.[147] In this latter study, the narrative texts were drafted by one of the investigators with input from a registered dietitian and reviewed by a Hispanic (target population) staff member for plausibility. The authors commented that this may more closely mimic the context in which these interventions are developed due to limited time and resources typically available for the development of health education interventions.

The objectives of this chapter are to: 1) describe the process we followed to develop the story-based intervention; 2) report the results of pilot testing; and, 3) discuss the questions and issues that arose during development. This information will be valuable for further work in the area of storytelling, as well as more broadly in terms of identifying and developing communication strategies for healthcare consumers.

2.2 Methods

The intervention was developed through a multi-staged process that began with a creative writer generating the stories. Parent experiences were based on the writer's interviews with a sample of families who attended the emergency department (ED) at Alberta Children's Hospital (ACH) with a child presenting with croup between April and September, 2005. The interviews were designed to recount the sequence of events from time of onset of symptoms through to post-ED follow-up, and to elicit the parents' emotional reaction to the experience including their perspectives on the ED management for their child. The creative

writer interviewed consenting parents during their ED stay and followed-up by telephone 10 to 14 days after the ED visit to obtain the parents' experiences following discharge.

Parents/caregivers were eligible if: 1) their child was 3 months to 6 years with a clinical diagnosis of croup who was assessed as being eligible for steroids as specified by the Alberta Medical Association Guidelines for croup; 2) they were fluent in English; 3) they were 18 years of age or older; and 4) they had a telephone and would be available for telephone follow-up 10 days after presenting to the ED. The process received ethics approval from the University of Alberta and the University of Calgary and institutional approval from the Alberta Children's Hospital in Calgary, Alberta and Stollery Children's Hospital in Edmonton, Alberta.

The five stories developed by the creative writer were reviewed by a convenience sample of 10 individuals with a variety of professional and personal backgrounds. The doctoral student (LH) revised and edited the stories based on the feedback and amalgamated the five stories into three. In addition, evidence for the natural history (e.g., signs and symptoms, symptom progression) and medical management (e.g., timing and route of epinephrine and dexamethasone administration) of croup and additional health information (e.g., how and when to contact a healthcare professional, when to seek emergency care) were incorporated either into the story or as part of the booklet. The three stories were reviewed for clinical accuracy by three ED physicians and a pediatric nurse.

While there are numerous formats and media to convey stories, a priori we chose to develop paper-based story booklets that could be given to parents in the ED. A graphic designer and illustrator created the format and layout and generated illustrations for the booklets in order to complement and enhance the stories. The format and illustrations were critiqued by the study investigators.

The three story booklets were tested through focus groups of parents for presentation, interest, style, and clarity. We identified parents for the focus groups through advertisements posted in numerous locations in Edmonton, including EDs, public health units, medical clinics, and local daycares. We initially aimed to recruit parents of children who had experienced croup in the previous year. Due to low numbers, we expanded the eligibility to include any parent with a young child (3-12 years old). The focus groups were conducted by a researcher with expertise in qualitative methods (Dr. Shannon Scott, Faculty of Nursing, University of Alberta). The focus groups ran for an hour and participants were reimbursed with $20 Canadian. During the focus groups, participants were encouraged to actively and creatively express their views in response to predetermined questions. Ethics approval for the focus groups was obtained from the University of Alberta prior to recruitment.

Finally, the booklets were presented at the 2007 Annual Meeting of the Pediatric Emergency Research Canada Network, a national organization of physicians and researchers from pediatric EDs across Canada. The main question for feedback was how to deliver the story booklets to parents, e.g., give all parents all booklets or target by severity of the child's illness.

2.3 Results

The creative writer generated five stories based on her interviews with the parents/caregivers of 10 children presenting to the ED. The five stories were designed to characterize different experiences and cover a range in terms of severity of illness and socioeconomic considerations (e.g., single mother, adolescent mother, Aboriginal background).

The initial feedback fell into four main categories: 1) overall concept; 2) format and presentation; 3) specific story content; and, 4) medical/health information. Reviewers generally liked the concept of the story booklets and found the project to be interesting and innovative. However, reviewers questioned the specific

purpose of the stories (e.g., comfort parents, impart knowledge) and the target audience (i.e., child versus parent).

Regarding the format, reviewers felt the stories were too long, the language was generally too advanced for an average reader, and some of the sentences were complex and awkward. Reviewers liked the illustrations but found the font too small. One reviewer found that the story titles were "blasé" and did not reflect the main content of the stories. Two reviewers wanted more dialogue or more of the characters' thoughts. Another reviewer commented on the homogeneity across the characters (e.g., parent, admitting physician, night nurse) and wanted more details about the characters and social context. One reviewer commented on the fact that the main character in each of the stories was the mother which may not accurately reflect contemporary parenting roles. Overall, the reviewers found the stories to be engaging largely due to the ability of the writer to capture the parents' emotions.

Several comments were made regarding specific aspects of the stories. For instance, one story described an infant having an x-ray:

> "Jimmy had never had an x-ray before, and Diane was not prepared for what she saw. Her heart broke as staff stripped her baby naked and strapped him onto a board which would hold him in place for the x-ray. Though the technicians were very careful with him, Diane was disturbed to see Jimmy crying in the brace. She knew the x-ray was important, but it was the hardest thing she had ever seen as a mom."

Three reviewers found this description to be too harsh and graphic; however, one reviewer thought it was important to prepare a parent for what they might experience. Some incongruencies were noted in the stories (e.g., a 13-month-old being transported in an infant car seat, distress about finding babysitting for a 13 year-old sibling). One reviewer couldn't identify with the main character from one of the stories and therefore didn't find the story engaging. One reviewer did not find the introduction to one of the stories captivating and therefore was not interested in reading on.

The final group of comments related to the medical and health information provided in the stories. Reviewers generally wanted as much information as possible about medical procedures and practices and considered the stories to be an excellent potential source of medical advice for parents. Reviewers wanted medical terminology to be explained (e.g., epinephrine mask, dexamethasone) and cautioned against inconsistent use of terminology (e.g., dexamethasone versus steroid).

Based on this feedback, the stories were substantially revised and reduced from five to three while capturing many of the events and the tone of the original stories. The revised stories were written using simpler language and sentence structure. The revised stories had a Flesch-Kincaid Grade Level Score of 6.2 indicating that a sixth grader (based on US school grade level) could understand them. The three revised stories each reflected a different severity of croup and different healthcare experiences: the mild case was managed at home; the moderate case was seen in the ED and discharged home; and, the severe case was hospitalized for two days. The main characters in the three stories reflected different demographics (e.g., married, single, male, female).

Eight individuals were involved in the focus groups. The results of the focus groups were categorized as: 1) general perceptions of the stories; 2) content and emotional by-products of the stories; 3) preferences; and, 4) graphics, layout, and illustrations. The focus group participants were generally very positive about the booklets; however, one participant expressed concern about the expense of the story booklets and potential wastefulness. The participants found the graphics and layout to be visually appealing: the booklets “caught my eye.” They found that they could identify with the stories: “the stories spoke to me.” They found that it was easier to get information from the stories compared to a typical information sheet from the ED. The participants suggested that the developer, or sponsor, of the story booklets be more visible to enhance credibility.

Regarding the content of the stories, the participants generally found the stories interesting, engaging, and easy to read. They found that the stories resonated with them and "matched" their personal experience. They found the information to be very helpful and that it "fit" the ED context. Further, they appreciated the suggestions in the stories, specifically how to cope with having a child with croup. The participants found the stories to provide comfort and emotional reassurance. However, the participants found it difficult to determine which of the story booklets would be most relevant for them (i.e., mild, moderate, or severe), and found the differences in terms of severity were not clear. They commented that explaining the rationale for treatments would be useful (e.g., why cold air helps). The participants highlighted some errors (e.g., use of term "web browser" rather than "search engine," inconsistencies in facts presented at the back of the books, typographical errors).

One issue relating to story content generated much discussion and consideration: in one of the stories, the parents did not take the child to the ED but managed the child's symptoms at home based on information they found through the internet. The participants suggested providing in the story booklets a list of recommended websites or information on how to evaluate websites and whether they are a trustworthy source of information. The participants also questioned whether receiving the internet story in the ED would be the most appropriate venue as the story was not relating the same circumstances as the other two stories. Participants felt that it may be better to receive such a story in a community health setting.

The focus group participants highlighted several preferences. The consensus for the most preferred story was "Things we take for granted" (Appendix D) as they found they could relate most to this story. They appreciated the fact that the story was written in the first-person mode and found that it held their attention better. The participants appreciated having a father as the main character in one of the stories and the fact that he accessed the internet for information. Finally, the participants liked the "catchy" titles of the stories, although some felt that the title should include "croup," or one or more of the symptoms.

The focus group participants found the presentation of the booklets soft and eye appealing. They enjoyed the variety in the illustrations (e.g., some full page, some half page). The participants all preferred the size of the booklet entitled "Things we take for granted;" they appreciated that it was the same shape and size as many children's books and saw that this was a positive feature. The participants enjoyed the illustrations and the colours used throughout the books; however, in one case the use of different coloured font for portions of the text created confusion for the reader regarding what was or was not important to read. The participants felt that the illustrations could appeal to both adults and children, thereby serving as a method for parents to explain to their child what may happen in the ED. Participants stressed that it would be important for the books to have a common format, shape, and size if there was going to be several series of these books (e.g., books on other disease/illness conditions).

We revised the story booklets based on the focus group feedback and presented the final products at a national conference of pediatric emergency researchers. The primary question for this group was how to package and disseminate the story booklets. The general consensus was to provide the three booklets to parents in a single package. We developed a folder for this purpose which held the three booklets. The final story booklets are presented in Appendix D.

2.4 Discussion

We followed a thorough and extensive process to develop the story booklets to provide information and comfort to parents attending the ED with a child with croup. We found that the development of such an intervention involved numerous decisions which were best informed through testing and refinement involving the end-user group. In general the feedback was very positive, although one focus group participant questioned the costs involved in producing the story booklets and whether the resources would be better spent elsewhere. A recurring theme of the feedback was the ability of the reader to relate to the stories and identify with the characters. In fact one reviewer said the stories brought tears to her eyes as she

recalled her own similar experiences. The specific feedback in terms of story content, errors and inconsistencies, and presentation style was critical for accuracy and to target the story booklets to the end-users' needs and preferences.

There were several challenges we encountered during the development of the story booklets. A key challenge, highlighted by one of the initial reviewers, was regarding the purpose of the story booklets. We hypothesized that the story booklets could serve a number of purposes, such as communicating information, contextualizing the illness experience and medical encounter, providing a decision aid, and building relationships between healthcare providers and consumers or among consumers undergoing the same experiences; however, our primary purpose was to provide information and comfort to parents. We considered this a critical initial step and would encourage others engaged in similar work to carefully consider the purpose of their intervention and what they plan to achieve through both product development and utilization. This is critical not only for the development of the interventions, but also to evaluate their effectiveness. The purpose of the intervention should be directly related to the outcomes to be assessed in its evaluation: e.g., communicating information (recall of information, satisfaction with information, compliance with information or instructions); contextualizing (comfort, anxiety); decision aid (decision regret, comfort/ease of decision-making, or subsequent resource use); and, building relationships (feelings of being supported, satisfaction).

A second challenge was staying true to the story versus being evidence-based. For instance, in one case the child was given an x-ray, despite the fact that this is not standard practice for croup and does not conform to accepted clinical practice guidelines. Our dilemma was whether to recount the events as described by this parent or reflect accepted clinical practice. In the end, we did not include the x-ray account and aimed to make the stories reflect typical cases of mild, moderate, and severe disease and how they would be managed on average. Another example was the situation where a child had been misdiagnosed prior to the ED visit. We had to consider whether it was appropriate to point out that physicians may make errors

in diagnosis. We decided to include the incident in the story to highlight a relatively common error of misdiagnosing croup for asthma. It also provided an opportunity in the story to educate the parents around the differences of croup and asthma, and particularly the different treatments appropriate for each.

Another dilemma regarding being evidence-based was whether or not to describe interventions for which there was no evidence. For instance, there is widespread practice and recommendations around the use of mist or humidity for croup despite no evidence supporting its effectiveness. A further issue was around the naming of drugs in the stories and the potential perception of product placement. For example, many parents would be more familiar with "Tylenol," rather than "acetaminophen." We chose to use the trade names that are more familiar to the lay person but included a range of product names to not appear preferential to a single brand.

A related issue was how much additional information or evidence to incorporate into the stories. Many of the reviewers wanted more detailed medical information; however, this was not typically captured in the parents' recounting of events. In the end, we included a fair amount of information about croup and its management (e.g., signs and symptoms, what is a steroid, how the drugs are administered), but tried to incorporate this detail as seamlessly as possible into the story while preserving its flow and tone. We also felt bound ethically to provide appropriate information on when to seek additional or emergency care. Further, based on feedback we added a foreward to each of the story booklets by the ED director of the local children's hospital as an endorsement of the story booklets by the healthcare system.

An issue that generated much discussion and controversy was the parents' reliance in one of the stories on information found on the internet. This raised concerns that we may inappropriately condone information that is found on the internet, as well as ethical concerns that we may be encouraging parents to manage their child's illness at home when the child may require medical care.

Nevertheless, we recognized that lay people are regularly using the internet as a source of medical information and felt that it was an important issue to profile in our stories. We addressed these concerns in several ways. First, we reviewed many of the websites that were identified when searching Google for "croup." Because the evidence is so strong for the management of croup, the information across websites was very consistent and accurate. Second, we suggested a website, that we know to provide reliable information, within the stories that parents could go to for additional information. Third, we included information at the end of each of the stories about when to seek medical care and specifically when to seek emergency care.

A major challenge was developing stories that would be widely generalizable and appealing. Numerous considerations arose such as how many stories, how long, reading level, narrative mode (e.g., first person, third person), representation of different demographics (e.g., sex, race, age, socioeconomic status), and representation of different illness experiences (e.g., severity of illness, hospitalization, management at home). We had to strike a delicate balance between being as inclusive, generalizable, and detailed as possible, while being as succinct as possible to increase the likelihood that parents would read the complete stories. These considerations need to be informed by the end-user group, specifically those who are most likely to benefit from the intervention.

A major consideration and investment of resources related to the presentation and packaging of the stories. There are many media through which stories can be delivered (e.g., computer, video, games, cartoons, etc). A priori we chose for this project to use paper-based story book formats. However, there remained many considerations within this format such as the type of illustrations, use of graphics and colour, and shape and size of the booklets. Again, the feedback we received through testing was helpful in making decisions. The format of delivering stories is particularly dependent on the preferences of the target end-user group.

While the feedback we gathered was rich and informative, our process was limited by the small number of individuals that we were able to recruit for our focus groups. We advertised widely across numerous venues and had very few willing participants. We found it particularly challenging to recruit as our target end-user was parents with young children who have many competing priorities and time constraints.

This chapter is being prepared to submit for publication. The authors and their contributions include: Lisa Hartling (project coordination, editing stories, data analysis and interpretation from initial testing, preparing manuscript); Shannon Scott (editing stories, conducting focus groups, analysis and interpretation of focus group data, preparing manuscript); Rena Pandya (project coordination, reviewing manuscript); Ted Bishop (reviewing and editing stories, reviewing manuscript); David Johnson (reviewing and editing stories, reviewing manuscript); Terry P. Klassen (reviewing and editing stories, reviewing manuscript). Other contributions included Jilleen Kosko (creative writer); Lara Minja and Matthias Reinicke from Lime Design Inc. (graphic designers); and, Val Lawton (illustrator).

Chapter 3

Methods for Randomized Controlled Trial

3.1 Trial overview

3.1.1 Trial Design: This was a randomized trial involving 2 sites: Stollery Children's Hospital (SCH) in Edmonton and Alberta Children's Hospital (ACH) in Calgary; both cities are in Alberta, Canada. Consenting parents of children with croup were randomized and received the intervention as early as possible during their ED visit (Appendix B – Information Sheets and Consent Forms). The parents were aware that the study was evaluating different approaches to managing children in the ED, but were not aware of the specific study hypotheses; they were assured that the study would not affect the medical management of their child. Because of the nature of the intervention, the research nurse and the other ED personnel involved in the study were not blind to the study groupings of the participants.

3.1.2 Participant follow-up: Parents were interviewed upon entry into the study and on discharge from the ED (Appendix C – Flow diagram of patient recruitment and follow-up); parents of children who were hospitalized remained in the study. Parents were contacted by telephone at 1 and 3 days following their visit to the ED. If the child had croup symptoms at the day 3 follow-up, the parents were contacted every two days until the symptoms resolved or up to day 9 post-ED visit.

3.2 Trial interventions

3.2.1 Experimental Intervention: The experimental intervention was three booklets that integrated stories, as told by parents of children with croup attending the ED, with evidence regarding the epidemiology and treatment of the condition. Each story reflected a case of different severity (mild, moderate, severe) which was clearly identified on the booklet's cover. The story booklets were given together in a folder specifically designed for the study.

3.2.2 Control Intervention: A standard information sheet produced by the Alberta Medical Association was the control intervention (Appendix E). The information

sheet describes what croup is, signs and symptoms, management, and when to consult medical services.

3.2.3 Timing of Intervention: The story booklets or standard information sheets were given to parents after they had been randomized to treatment groups (i.e., as early as possible during their ED visit). The intent of this timing was to provide the opportunity for them to peruse the information during their ED stay.

3.2.4 Reading Levels: The story booklets and information sheet both had a Flesch-Kincaid Grade Level Score of 6.2 indicating that a sixth grader (based on U.S. school grade level) could understand the documents.

3.2.5 Co-Interventions: Parents and their children received usual care in addition to the story booklets or information sheet. Part of that care involves communication with the health professionals in the ED. It was expected that randomization would help to balance the groups with respect to any co-interventions.

3.3 Allocating participants to trial groups

The doctoral student (LH) prepared the randomization sequence using Microsoft Excel 2003 and prepared the sealed, opaque envelopes. After obtaining informed, written consent from the parent, the research nurse/assistant opened the next envelope in a series of consecutively labeled, sealed, opaque envelopes. These were kept in a secured location in the ED. The research nurse/assistant and treating physician were unaware of the next group assignment.

3.4 Methods for protecting against sources of bias and lack of precision

3.4.1 Blinding: Parents were blind to the interventions being compared. While they were aware that the study was evaluating some aspect of the management of children with croup and their families, they did not know what aspect of management was being tested. Because of the nature of the intervention, the research nurse/assistant and other ED personnel were not blind to the intervention

that participants received. Our intent was to blind those conducting the follow-up interviews to the intervention the participants received by having different individuals performing recruitment and follow-up; however, this was not always possible due to logistics of staffing and funding.

3.4.2 Contamination: There was a possibility for contamination if parents in one study group were exposed to the intervention given to the other study group. Evidence shows that contamination is generally overestimated.[65] The consequence of contamination is to dilute the treatment effect, therefore the planned sample size was adjusted to account for this potential (Section 3.10). Contamination was assessed by asking parents if they received any other information related to croup.

3.4.3 Outcome measurement: Where possible, pre-existing tools that have established reliability and validity were used to assess outcomes (Section 3.8). Where such psychometric properties were not available, we used outcome measurement strategies that have previously been used and documented in the literature.

3.5 Inclusion and exclusion criteria

Parents of children with, or suspected of having, a clinical diagnosis of croup were eligible for study. Parents had to meet the following additional criteria: 1) have a telephone and be willing to be contacted for follow-up interviews; 2) fluent in English; 3) provide informed consent; 4) no prior visit to an ED during this episode of the disease; 5) no prior visit to an ED for another episode of croup during the study period. Parents were excluded if: 1) stridor was due to another cause (e.g., bacterial tracheitis, presence of a supraglottic foreign body); 2) parent had previously been included in the study.

3.6 Duration of treatment period

The proposed duration of recruitment/intervention was from October 1, 2007 until the required sample size of 420 was achieved. Initially, we estimated that we could achieve this sample size across the two sites during one croup season

ending March 31, 2008. Due to logistical impedances, we still had not recruited the full sample size after a second season (October 1, 2008 to March 31, 2009). The analysis presented in this document is based on the 255 participants that had been enrolled up to March 31, 2009; this represents 61% of the initial sample size projections.

3.7 Frequency and duration of follow-up

The research nurse/assistant in the ED recruited participants and obtained informed consent. Immediately after consent, the research nurse/assistant collected demographic information and participants completed a questionnaire on anxiety (Section 3.8.1). The research nurse/assistant assessed the severity of the child's illness using the Westley Croup Score (Appendix F – Baseline interview, Part E).[165] The research nurse then opened the next allocation envelope and documented which intervention the parent received. On discharge from the ED, participants completed another short questionnaire to assess parental anxiety. The research nurse/assistant contacted the parent at 24 hours (1 day) and 3 days after the ED visit. Parents of children who were still symptomatic at day 3 were contacted every 2 days until the symptoms resolved or until day 9.

3.8 Primary and secondary outcome measures

3.8.1 The primary outcome was **change in parental anxiety** from baseline (immediately following recruitment to the study) to discharge from the ED. State anxiety (of interest here) refers to emotional reactions characterized by subjective, conscious feelings of tension, apprehension, nervousness, and worry.[7] This was measured using the Spielberger State-Trait Anxiety Inventory which is a well-known instrument designed to measure state anxiety at the time of administration, in the recent past, or at a future point in time (STAI-S, Form Y). The inventory consists of 20 items that ask respondents to indicate how much each statement reflects how they feel on a 4-point Likert scale ranging from "not at all" to "very much so" (Appendix F – Baseline interview, Part A). Scores are summed; the range of possible scores is 20 to 80. Higher scores indicate higher anxiety. The

scale has good internal consistency and takes 6-10 minutes to complete during initial administration and less than 5 minutes during repeat administrations.[151]

*3.8.2 Secondary outcomes**:***

i) **Expected future anxiety**: The STAI-S was administered at 1-day post-visit to gather self-reports of expected anxiety should they face another incident with croup in the future.

ii) **Event Impact**: The Impact of Event Scale (Appendix F – Day 3, 5, 7, 9 Telephone Interview, Part B) includes 15 self-report items to measure intrusion (7 items) and avoidance (8 items) resulting from exposure to anxiety-producing events (in this case, the child's croup illness). Using a 4-point Likert scale ranging from "not at all" to "often," respondents indicated how frequently the items were relevant to them during their child's illness with croup. This tool has been shown to have good internal consistency and takes up to 10 minutes to complete.[75] This scale was administered during the last telephone follow-up (i.e., when the child was symptom-free).

iii) **Parental knowledge** about the natural history of the disease, symptoms, and management strategies were assessed using questions that were developed specifically for this study (Appendix F – Day 3 Telephone Interview, Part C). The questions were based on information that appears in the story booklets and the information sheets. The knowledge assessment tool has not been validated, nor are there any gold standard scales in the literature. The questions were evaluated for face validity by the investigative team. The questions were pilot tested for clarity among a sample of 7 parents prior to commencement of the trial. The knowledge questions were asked at 3 days post-visit to assess short-term recall.

iv) **Parental satisfaction** with the overall ED visit and the information they received was assessed using independent questions with responses on a 5-point Likert scale ranging from excellent to poor (Appendix F – Day 1 Telephone

Interview, Part D). These questions were developed specifically for this study based on evidence from the literature.[158] This was assessed at 1-day post-ED visit.

v) **Parental decisional regret** (i.e., "remorse or distress over a decision")[16] regarding the decision to take their child to the ED was assessed using a validated scale.[16] The tool has parents rate five statements from strongly agree to strongly disagree (Appendix F – Day 1 Telephone Interview, Part C). This outcome provides indirect evidence of the parent's satisfaction with bringing their child to the ED. This was assessed at the 1-day telephone interview.

vi) **Incidence of return to be evaluated by a physician (or other health care practitioner) for croup** was assessed throughout the follow-up interviews.

vii) **Healthcare utilization patterns**: Parents were asked whether they sought further medical care for this episode of croup following the visit to the ED. If they answered "yes," they were asked about the type of consultation (e.g., in-person, telephone health advice service), location of care, type of care provider, and whether they were prescribed any medication.

viii) **Resource utilization:** Family use of resources was assessed during follow-up interviews through questions regarding costs for medication, equipment (e.g., humidifiers), parking and travel, ambulance service, child care, and time lost to usual activities. Information on costs was collected at each of the follow-up interviews.

ix) **Ongoing croup symptoms** were assessed using the Telephone Outpatient Score for Clinical Status (TOP score; Appendix F – Day 1 Telephone Interview, Part B).[13,83] The TOP score involves three questions dealing with croup symptoms. These were assessed at each telephone follow-up. This information was collected to compare groups with respect to the course of the disease.

3.9 Outcome measurement at follow-up

3.9.1 In-person Interviews: Immediately following consent, the research nurse/assistant administered a questionnaire to gather demographic information and the parent completed the STAI-S to document baseline anxiety. If both parents accompanied the child, we asked the primary caregiver to participate or the secondary caregiver if the primary caregiver was unwilling. If both parents shared equal caregiving responsibilities, we asked them to choose who would participate; however, we asked that the same parent complete all of the follow-up interviews for consistency. On discharge from the ED, the same parent was asked to complete the STAI-S again.

3.9.2 Telephone Interviews: Other outcomes listed in Section 3.8.2 were measured through telephone interviews conducted at 1, 3, 5, 7, and 9 days following the ED visit. A trained individual administered the questionnaires (Appendix F) using standardized telephone interviewing techniques.

3.10 Planned sample size

The alternate hypothesis was that change in anxiety from beginning to end of the ED visit would be different for the group receiving story booklets versus the comparison group. In the absence of data specific to parents of children with croup attending the ED, the estimates for sample size calculations were based on previous research in similar clinical populations. The initial anxiety level in both groups was estimated to be approximately 45 on the STAI-S scale.[11,24,46,53,69,76,87,119,167] Two studies of parents whose children were undergoing elective surgery reported anxiety levels of 45.97 and 44.76, respectively.[24,49] We believed these estimates to be conservative; for instance, parents bringing their young (<2 years) febrile children to the ED scored 50.1;[129] parents of hospitalized children requiring total parenteral nutrition showed baseline anxiety levels of 59.5;[101] and parents of children admitted to the pediatric intensive care unit showed levels of 52.8.[119] In the latter study evaluating a multi-faceted intervention including storytelling, mothers in the intervention and control groups had average

anxiety levels of 36 and 40, respectively, one-month post-discharge. Based on these findings, we hypothesized that parents in the story booklet group would return to a "normal" level of anxiety following treatment (i.e., 36 or 37),[151] while those in the comparison group would remain more anxious (i.e., 39 or 40).[151] We conducted sample size calculations, using a two-sided, two sample t-test with a significance level of 0.05 and standard deviation of 10 (based on the cited studies), to detect a difference of 3 or 4 points on the STAI-S scale. This effect size (0.3 and 0.4 respectively) is comparable to previous research evaluating an intervention involving a story [119] and written information provided to parents of hospitalized children.[101] For 80% power, we required 100 or 176 individuals per group for a 4 or 3-point difference, respectively. The sample size was inflated by 20% (210 per group) to account for potential contamination and drop-outs.[65] We conducted power calculations for difference in parental knowledge between groups, as this secondary outcome may lie in the causal pathway of provision of information and anxiety reduction. With 210 participants per group, we would have 99% power to detect a moderate (0.5) or large (0.8) effect size, and 80% power to detect an effect size of 0.28 (where 0.2 is considered small).[35] For the current analysis of 255 patients, we have 80% power to detect an effect size of 0.35.

3.11 Recruitment procedures

3.11.1 Recruitment Rate and Time Period: Recruitment was planned to take place from October 1, 2007 until the required sample size was achieved, or until March 31, 2008. The incidence of croup varies biannually with the highest rates occurring during the season beginning in an odd numbered year (e.g., September 2007).[113] Based on previous experience, we anticipated a recruitment rate of between 65% and 85% [13,93,94,124] of all patients who were assessed for eligibility. Based on ED utilization data, there were 1,640 cases seen at the ACH and SCH between October 2003 and March 2004 which would result in 1,066 and 1,394 cases based on recruitment rates of 65% and 85%, respectively. We planned to recruit 210 parents per group to have adequate power and to allow for some loss

to follow-up over the study period. Despite this detailed planning, we were unable to recruit the anticipated sample size in the proposed time period.

*3.11.2 Recruitment Process**:*** Parents of children in the ED with suspected croup were identified by the triage nurse, or other staff nurse, who notified one of the study personnel. Study personnel were on-site during the evenings, primarily from 6:00 to midnight, which corresponds to the period with the highest number of croup visits. The research nurse/assistant approached the parents and explained the study and invited them to participate. After obtaining written, informed consent (Appendix B), the research nurse/assistant assessed and documented the severity of the child's condition, and administered the baseline questionnaire to determine study eligibility. If eligible for enrolment, the research nurse provided the participant with the experimental or control intervention based on their treatment allocation. On discharge, the research nurse or research assistant documented the patient's disposition. We maintained a register of eligible participants who refused, were missed, or were otherwise excluded.

3.12 Compliance: Parents were given the interventions, but it was their choice to read the information. To assess compliance, parents were asked during the follow-up interviews whether they read the information they were given. All parents were included in the analysis regardless of whether or not they read the intervention material.

3.13 Data analysis

*3.13.1 Baseline variables**:*** Baseline variables were described for each group overall and by study site. Imbalances between intervention groups for key baseline variables were noted.

*3.13.2 Primary outcome**:*** For self-reported anxiety, a change score from baseline to discharge was calculated for each patient. The median change scores were compared between groups using the Mann-Whitney test.

3.13.3 Secondary outcomes: Continuous outcomes (e.g., knowledge, decisional regret) were compared between study groups using independent-groups t-tests if the data were normally distributed, and the Mann-Whitney test if the data were skewed. Categorical outcome data (satisfaction) were analyzed using the Chi-square test. Kaplan-Meier curves for time to resolution of symptoms were tested for equality using the log rank (Mantel-Cox), Breslow (Generalized Wilcoxon), and Tarone-Ware tests.

3.13.4 Analytic approach: Our primary analysis was based on intention-to-treat approach where all participants who were randomly assigned to a study group were included whether or not they received or complied with (i.e., read) the intervention to which they were assigned. Statistical analyses were conducted using statistical software SPSS (version 17; SPSS Inc. Chicago, IL). The significance level was set at 0.05.

3.13.5 Subgroup analyses: All analyses were performed separately by site (ACH, SCH) to identify any differences in the pattern of results.

3.14 Ethical considerations

Study participation presented no known risks, inconvenience, cost, pain or suffering to the participants (parents and pediatric patients). Patients received standard medical management at the discretion of the attending physician. The study was approved by the Ethics Review Boards at the University of Alberta and the University of Calgary prior to commencement. Given that the study intervention did not pose any direct risk to patients or health care professionals, a Data Safety and Monitoring Committee was not necessary.

The content of this chapter formed the basis of a proposal submitted for funding to the Canadian Institutes of Health Research. The following individuals contributed to various aspects of the study design and reviewed the proposal prior to submission and study implementation: Lisa Hartling (overall coordination, proposal development, and writing), Shannon Scott (qualitative methods), David

Johnson (clinical expertise, quantitative methods), Ted Bishop (creative writing), Jamie Brehaut (cognitive psychology), Gillian Currie (economic analysis), Ben Vandermeer (statistical analysis), Mandi Newton (outcome measures related to mental health), and Terry P. Klassen (clinical expertise and study conception).

Chapter 4

Results of Randomized Controlled Trial

4.1 Study sample

Overall 255 parents were recruited: 129 participants were randomized to receive story booklets and 126 received standard information sheets. Figure 4.1 describes the recruitment and follow-up of study participants to day 3 which was the last follow-up point required for all participants.

Characteristics of the trial participants are detailed in Tables 4.1 to 4.3. There were no notable differences between groups in terms of demographic variables (Table 4.1). There were 137 participants from ACH and 118 from SCH. The results by site are presented in Appendix G. The focus of this chapter is on the overall results (both sites combined) and only presents site-specific results if they differ from the overall results.

Table 4.2 presents the results for parental concern at baseline. Overall, parents demonstrated a moderate level of concern with a mean self-rating of 6.3 (SD 2.5) on a scale of 1 to 10, where 10 represents the highest level of concern. The items that generated the most concern were the unusual sound of the child's breathing (40.4% expressed extreme concern) and the effort the child made to breathe (42.7% extreme concern). There were no notable differences in overall or item-specific concern between study groups.

The majority of participants had no prior history of croup admissions, ICU admissions, or intubations. A substantial proportion (>40%) of participants reported a previous experience with croup either with the same or another child, while 23% reported a prior serious illness or medical condition for their child. The most commonly reported serious illnesses/medical conditions were asthma (n=26), pneumonia (n=7), and complications of prematurity (n=6). Overall there were no differences between groups in the prevalence of previous medical history or experiences with participants' children.

The majority of patients presented with mild croup with a median croup score of 2 (IQR 1,3) on a scale of 0 to 17. Approximately 90% of the patients were

discharged home from the ED with less than 5% being admitted. Approximately 1 in 5 children had been seen by the staff physician before being recruited into the study. Further, treatment had already been ordered for almost 70% of patients prior to recruitment. The care prior to recruitment differed between the two sites. All of the patients at ACH were seen by a triage nurse prior to recruitment and 87% had been ordered treatment. At SCH, a greater proportion had been seen by the staff physician but only half had been ordered treatment prior to recruitment.

Approximately 60% of participants read the study material during their ED stay while 20% read additional information on croup. The groups differed somewhat with fewer parents in the story group reading the study material (53% versus 66%) but more parents in the story group reading additional material (33% versus 17%).

Figure 4.1. Recruitment and follow-up of study participants

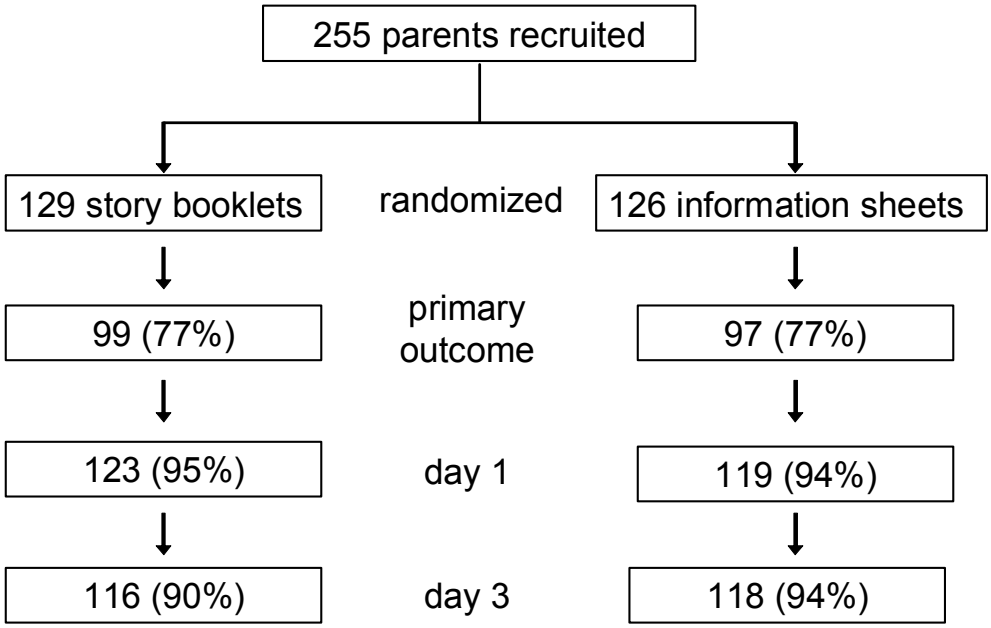


Table 4.1 Demographics

	Story booklets		Information sheet	
	N		N	
	129		**126**	
Age of participant, years (mean, SD)	34.6	5.92	32.9	5.99
Sex of participant				
Female	99	76.7%	102	81.0%
Male	29	22.5%	21	16.7%
Unknown	1	0.8%	3	2.4%
Sex of child				
Female	50	38.8%	40	31.7%
Male	78	60.5%	86	68.3%
Age of child, years (median, IQR)	2.12	1.08,3.92	1.92	1.17,3.23
Site				
Alberta Children's Hospital	70	54.3%	67	53.2%
Stollery Children's Hospital	59	45.7%	59	46.8%
Number adults living in the home				
1	8	6.2%	11	8.7%
2	108	83.7%	96	76.2%
>2	12	9.3%	19	15.1%
Number adults participating in care of child				
1	5	3.9%	8	6.3%
2	104	80.6%	96	76.2%
>2	19	14.7%	22	17.5%
Total number of children living in the home (median, IQR)	2	1,3	2	1,3
Relationship to child				
Parent	125	96.9%	123	97.6%
Other	2	1.6%	2	1.6%
Education				
grades 1-9	1	0.8%	0	0.0%
grades 10-11 (some high school)	5	3.9%	5	4.0%
high school graduate	20	15.5%	31	24.6%
some college/university	21	16.3%	27	21.4%
college graduate	42	32.6%	39	31.0%
post-graduate education or degree	37	28.7%	21	16.7%
Marital status				
never married	5	3.9%	11	8.7%
married/common-law	114	88.4%	102	81.0%

separated, divorced, or widowed	7	5.4%	11	8.7%
other	1	0.8%	1	0.8%
Household income (Cdn $)				
<15,000	5	3.9%	3	2.4%
15-29,000	5	3.9%	7	5.6%
30-44,000	8	6.2%	8	6.3%
45-59,000	11	8.5%	13	10.3%
60-74,000	14	10.9%	12	9.5%
75-90,000	10	7.8%	19	15.1%
>90,000	61	47.3%	47	37.3%
NR	15	11.6%	17	13.5%
Ethnic or minority group				
No	99	76.7%	92	73.0%
Yes	23	17.8%	28	22.2%
Place of birth				
North America	96	74.4%	101	80.2%
Outside of North America	29	22.5%	21	16.7%

SD=standard deviation; IQR=inter-quartile range

Table 4.2 Parental concern at baseline

	Story booklets		Information sheet	
	N		N	
	129		126	
Level of concern about the following items:				
uncomfortable aspect of child's cough				
0 (not at all)	5	3.9%	1	0.8%
1	28	21.7%	22	17.5%
2	52	40.3%	56	44.4%
3 (extremely)	43	33.3%	47	37.3%
NR	1	0.8%	0	0.0%
unusual sound or nature of the cough				
0 (not at all)	8	6.2%	2	1.6%
1	25	19.4%	30	23.8%
2	47	36.4%	39	31.0%
3 (extremely)	48	37.2%	55	43.7%
NR	1	0.8%	0	0.0%
unusual sound of child's breathing				
0 (not at all)	7	5.4%	5	4.0%
1	20	15.5%	15	11.9%
2	45	34.9%	41	32.5%
3 (extremely)	57	44.2%	65	51.6%
NR	0	0.0%	0	0.0%
effort that child is making to breathe				
0 (not at all)	12	9.3%	10	7.9%
1	24	18.6%	25	19.8%
2	38	29.5%	37	29.4%
3 (extremely)	55	42.6%	54	42.9%
NR	0	0.0%	0	0.0%
child is not getting enough oxygen				
0 (not at all)	25	19.4%	21	16.7%
1	32	24.8%	28	22.2%
2	40	31.0%	41	32.5%
3 (extremely)	31	24.0%	36	28.6%
NR	1	0.8%	0	0.0%

child may be wheezing or have asthma				
0 (not at all)	29	22.5%	25	19.8%
1	19	14.7%	30	23.8%
2	46	35.7%	33	26.2%
3 (extremely)	34	26.4%	37	29.4%
NR	1	0.8%	1	0.8%
child's sleep was disturbed				
0 (not at all)	15	11.6%	14	11.1%
1	23	17.8%	20	15.9%
2	43	33.3%	42	33.3%
3 (extremely)	48	37.2%	50	39.7%
NR	0	0.0%	0	0.0%
parent felt increasingly tense or frustrated as a result of the illness				
0 (not at all)	27	20.9%	18	14.3%
1	27	20.9%	32	25.4%
2	41	31.8%	50	39.7%
3 (extremely)	34	26.4%	26	20.6%
NR	0	0.0%	0	0.0%
child might be hospitalized				
0 (not at all)	27	20.9%	28	22.2%
1	37	28.7%	44	34.9%
2	39	30.2%	28	22.2%
3 (extremely)	26	20.2%	25	19.8%
NR	0	0.0%	1	0.8%
illness might recur in the future				
0 (not at all)	10	7.8%	12	9.5%
1	31	24.0%	34	27.0%
2	42	32.6%	34	27.0%
3 (extremely)	46	35.7%	46	36.5%
NR	0	0.0%	0	0.0%
not knowing about this illness				
0 (not at all)	20	15.5%	25	19.8%
1	36	27.9%	41	32.5%
2	36	27.9%	31	24.6%
3 (extremely)	35	27.1%	27	21.4%
NR	2	1.6%	2	1.6%
Overall concern (scale 1-10) (mean, SD)	6.10	2.58	6.48	2.36

NR=no response; SD=standard deviation

Table 4.3 History of previous illness, severity of illness at baseline, and ED visit

	Story booklets		Information sheet	
	N		**N**	
	129		**126**	
History				
Parent first noticed respiratory symptoms (number of days to ED visit) (median, IQR)	1	0,2	1	1,2
prior history of croup				
no history	69	53.5%	64	50.8%
history same child	26	20.2%	23	18.3%
history other child	19	14.7%	20	15.9%
history both	13	10.1%	18	14.3%
prior history of croup admissions				
no admits	112	86.8%	102	81.0%
ED visit only this child	6	4.7%	7	5.6%
ED visit only other child	2	1.6%	6	4.8%
previous admissions this child	4	3.1%	4	3.2%
previous admissions other child	4	3.1%	6	4.8%
prior admissions to ICU				
no ICU admits	126	97.7%	121	96.0%
ICU this child	0	0.0%	2	1.6%
ICU other child	1	0.8%	1	0.8%
prior intubations				
no history	114	88.4%	110	87.3%
history this child	8	6.2%	7	5.6%
history other child	6	4.7%	6	4.8%
history both	0	0.0%	2	1.6%
prior serious illness or chronic medical condition this child				
No	101	78.3%	93	73.8%
Yes	27	20.9%	32	25.4%
Croup severity				
total score (median, IQR)	1	0,3	2	1,3
0	38	29.5%	24	19.0%
1	30	23.3%	31	24.6%
2	18	14.0%	27	21.4%
3	19	14.7%	22	17.5%
4	12	9.3%	15	11.9%
5	7	5.4%	4	3.2%
>5	4	3.2%	3	2.4%

missing	1	0.8%	0	0.0%
ED Care				
Disposition				
left without being seen	1	0.8%	4	3.2%
discharged home	116	89.9%	113	89.7%
Admitted	6	4.7%	7	5.6%
Other	6	4.7%	2	1.6%
Prior to recruitment patient seen by				
triage nurse	123	95.3%	122	96.8%
staff nurse	57	44.2%	58	46.0%
Resident	18	14.0%	23	18.3%
staff physician	28	21.7%	24	19.0%
Other	5	3.9%	6	4.8%
Prior to recruitment treatment ordered				
Yes	87	67.4%	87	69.0%
No	34	26.4%	34	27.0%
Read information during ED visit				
Read study material	68	52.7%	83	65.9%
Read additional information	31	24.0%	19	15.1%

ED=emergency department; IQR=interquartile range; ICU=intensive care unit

4.2 Primary Outcome: change in parental anxiety from baseline to discharge

The baseline anxiety score on the STAI was 37.2 (SD 12.3) for the story group versus 38.8 (SD 12.3) for the comparison group (Table 4.4). At discharge the STAI scores were approximately 5 to 6 points lower for both groups (32.2 and 32.8, respectively). There was no significant difference between groups in change in parental anxiety from baseline to discharge (p=0.78).

4.3 Secondary Outcomes

4.3.1 Expected future anxiety: The expected future anxiety as measured by the STAI during the Day 1 telephone follow-up showed no significant differences between groups (42.0 versus 42.6, p=0.36). Interestingly, the expected future anxiety was substantially higher than the participants' baseline anxiety (Table 4.1).

4.3.2 Event Impact: The impact resulting from exposure to anxiety-producing events was measured during the last telephone follow-up; this varied from day 3 to day 9 depending on when symptoms resolved. There were no significant differences between groups either overall (median=9 for both groups, p=0.912) or for the two subscales: intrusion (median=6 for both groups, p=0.945) and avoidance (median 3 versus 3.5, p=0.998).

4.3.3 Parental knowledge: There was no significant difference in knowledge between the two groups during the day 3 follow-up (8.57 versus 8.44, p=0.5). Overall, the knowledge level was high for both groups with a mean of 8.5 (SD 1.45) out of 10.

4.3.4 Parental satisfaction: The majority of patients in both groups (64% and 68% respectively) were "very satisfied" with the treatment and care they received in the ED. A further 19% and 21%, respectively, were "somewhat satisfied." The results for satisfaction around their expectations for information were similar with the majority "very satisfied" (77% and 71%) or "somewhat satisfied" (17% and 21%). There was no significant difference between groups in satisfaction with

respect to the participants' expectations for treatment and care or their expectations for information.

4.3.5 Parental decisional regret: The mean regret score, assessed at 1 day post-ED visit, was higher in the story group compared to the comparison group (1.26 versus 1.15). The difference between groups was statistically significant (t-test, $p<0.001$). When the five items in the regret scale were assessed independently, only one item showed a significant difference between groups (Table 4.5). More parents in the story group showed less agreement with the statement "I would go for the same choice if I had to do it again" ($p=0.017$). When analyzed by site, the same pattern of significance held for SCH; however, the results were not statistically significant for ACH.

4.3.6 Incidence of return to be evaluated by a physician (or other health care practitioner) for croup: More participants in the story group returned to a physician or the ED compared to the comparison group; the difference was not statistically significant (30.3% versus 24.8%, $p=0.334$).

4.3.7 Healthcare utilization patterns: There were no significant differences between groups in the incidence of contacting a healthcare professional following the ED visit (32.8% story group versus 26.4% comparison group). The most commonly contacted health professional was doctors (32.8% story group versus 41.7% comparison group, $p=0.153$), followed by return to ED (8.2% story group versus 6.6% comparison group, $p=0.637$), HealthLink (4.9% story group versus 3.3% comparison group, $p=0.527$), and other health professional (1.6% story group versus 0% comparison group, $p=0.157$). The two other health professionals contacted were a homeopath and a registered nurse. When analyzed by site, there were significant differences for SCH with the story group making more contacts with healthcare professionals overall (34.0% versus 17.0%, $p=0.045$), as well as contacts with HealthLink specifically (7.5% versus 0%, $p=0.038$).

4.3.8 Resource utilization: No participants used an ambulance following enrollment in the study. One child in the story group was hospitalized after being discharged home from the ED. Ten participants in the story group obtained prescription medications after being discharged from the ED compared to 13 participants in the comparison group. Most often prescribed was dexamethasone (n=15), followed by ventolin (n=3), amoxicillin (n=4), zithromax (n=1), prednisone (n=1), Q-var (n=1), Advair (n=1), and motrin (n=1).

4.3.9 Ongoing croup symptoms: Median number of days to no symptoms (TOP score=0) was 3 (IQR 3,5; SE=0.154) for the story group and 5 for the comparison group (IQR 3,5; SE=0.186). The survival distributions for the two groups were significantly different based on the log rank (Mantel-Cox) test (p=0.032) and of borderline significance based on the Breslow (Generalized Wilcoxon) (p=0.066) and Tarone-Ware (p=0.051) tests (Figure 4.2). The same pattern of results was observed when analyzed by site for ACH (p=009, log rank; p=0.057, Breslow; p=0.028, Tarone-Ware). Results were not statistically significant for SCH.

Table 4.4 Comparison of primary and secondary outcomes

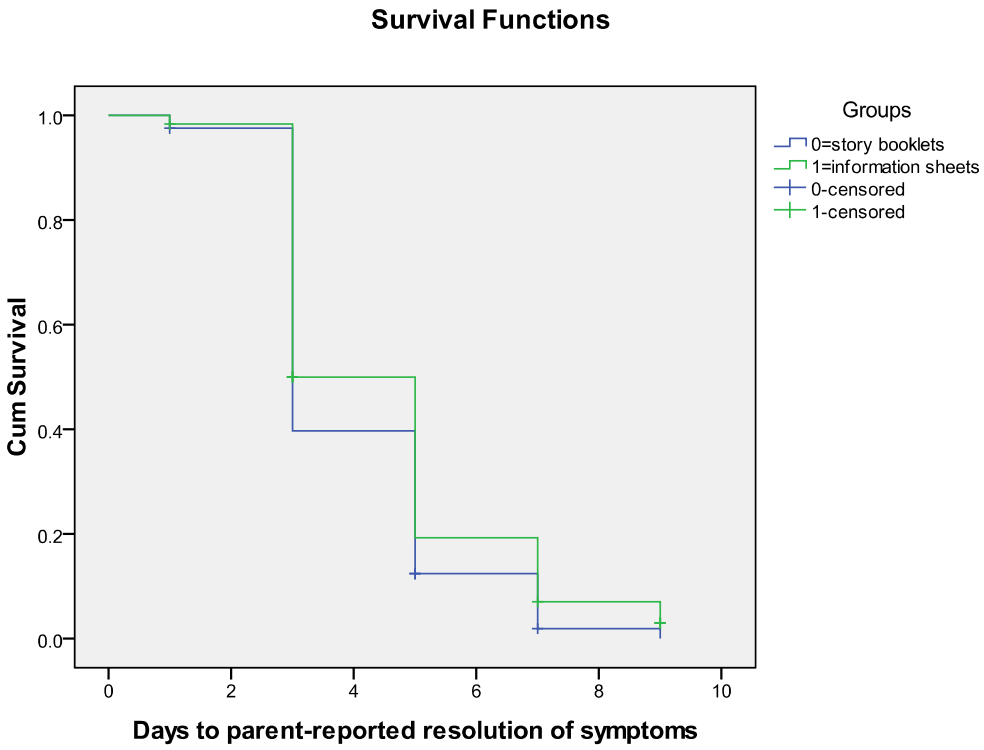
	Story booklets	Information sheets	P-value
Anxiety - STAI			
Baseline (mean, SD)	37.6 (12.3)	38.8 (12.3)	0.22
Discharge (mean, SD)	32.2 (11.1)	32.8 (9.72)	0.72
Discharge-Baseline (median, IQR)	6 (2,11)	7 (2,13)	0.78
Expected anxiety in future episodes of croup (measured at day 1 or 3 post-ED visit) (mean, SD)	42.0 (12.7)	42.6 (11.9)	0.36
Decision Regret (measured at day 1 or 3 post-ED visit)			
mean (SD)	1.26 (0.45)	1.15 (0.27)	p<0.001
Satisfaction (Day 1 or 3)			
Expectations for treatment and care (n)			p=0.21
very satisfied	83 (68%)	86 (72%)	
somewhat satisfied	25 (20%)	29 (24%)	
neither satisfied nor dissatisfied	4 (3%)	1 (1%)	
very dissatisfied	5 (4%)	4 (3%)	
NR	5 (4%)	0 (0%)	
Expectations for Information (n)			p=0.21
very satisfied	97 (80%)	89 (74%)	
somewhat satisfied	21 (17%)	27 (23%)	
neither satisfied nor dissatisfied	2 (2%)	1 (1%)	
very dissatisfied	0	3 (3%)	
NR	2 (2%)	0	
Knowledge (Day 3) mean (SD)	8.57 (1.59)	8.44 (1.30)	p=0.50
Impact of event scale (Last follow-up))			
Intrusion sub-scale (median, IQR)	6 (2,11)	6 (1,11)	p=0.95
Avoidance sub-scale (median, IQR)	3 (0,7.75)	3.5 (0,6.25)	p=0.99
Total (median, IQR)	9 (3,18.5)	9 (3.75,20)	p=0.91

STAI=State Trait Anxiety Inventory; IQR=interquartile range; SD=standard deviation; ED=emergency department; NR=not reported

Table 4.5 Comparison of decision regret scale

	Story booklets	Information sheets	P-value
	N=122	N=120	
Decision Regret (measured at day 1 or 3 post-ED visit): Parents were asked to respond to the questions regarding their decision to take their child to the ED for the episode of croup in question.			
It was the right decision.			p=0.405
Strongly agree	90 (73.8)	97 (80.8)	
Agree	28 (23.0)	20 (16.7)	
Neither agree nor disagree	1 (0.8)	2 (1.7)	
Strongly disagree	3 (2.5)	1 (0.8)	
I regret the choice that was made.			p=0.495
Strongly agree	2 (1.6)	0	
Agree	0	1 (0.8)	
Neither agree nor disagree	6 (4.9)	4 (3.0)	
Strongly disagree	113 (92.6)	114 (95.0)	
I would go for the same choice if I had to do it over again.			p=0.017
Strongly agree	79 (64.8)	95 (79.2)	
Agree	25 (20.5)	17 (14.2)	
Neither agree nor disagree	7 (5.7)	4 (3.3)	
Strongly disagree	11 (9.0)	2 (1.7)	
The choice did my child a lot of harm.			p=0.189
Strongly agree	0	0	
Agree	2 (1.6)	1 (0.8)	
Neither agree nor disagree	3 (2.5)	0	
Strongly disagree	117 (95.9)	119 (99.2)	
The decision was a wise one.			p=0.388
Strongly agree	96 (78.7)	94 (78.3)	
Agree	24 (19.7)	25 (20.8)	
Neither agree nor disagree	0	1 (0.8)	
Strongly disagree	2 (1.6)	0	

Figure 4.2. Comparison of survival functions for time to no symptoms: story booklets (group 0) versus standard information sheet (group 1)



Chapter 5

Discussion of Randomized Controlled Trial

5.1 Overview of Findings

This is one of few randomized controlled trials to examine the use of stories or narratives to communicate with healthcare consumers. The consumers of interest in this study were parents or caregivers of children attending the ED with croup. We compared parent and child outcomes following the ED visit for groups receiving story booklets versus a standard information sheet. Both interventions provided factual information about the signs and symptoms of croup, medical management, and when to seek medical and/or emergency care. The story booklets couched the information within real parent stories.

We found no significant difference for the primary outcome of change in anxiety between the time of study enrolment and discharge from the ED. We found several differences in outcomes following the ED visit including more decision regret and quicker time to resolution of symptoms for the story group. There were no significant differences for the remaining outcomes.

The parents who received the story booklets showed greater decision regret compared to the parents who received the standard information sheets. The measurement scale we used was developed to measure regret in healthcare decisions at a given point in time.[16] The parents in our study were asked to respond to the tool with respect to their decision to take their child to the ED for the episode of croup in question. The greater decision regret could be interpreted in at least two ways. First, parents may have regretted their trip to the ED because of the care or information they received. This explanation is less likely as there were no differences between groups in satisfaction with care or information received in the ED. Alternatively, after reading the story booklets parents may have felt that they could have managed the child at home and avoided the trip to the ED. This explanation may be partly substantiated by the fact that the single item driving the difference in decision regret was parents' response to the question, "I would go for the same choice if I had to do it over again," with parents in the story group showing less agreement with the statement. Further

research is required to corroborate this finding, as the significant results may have arisen due to chance, and to understand the mechanism that might create different levels of decision regret in this context. While the difference between groups in decision regret was statistically significant, the absolute difference is of questionable clinical or practical importance. Overall, the decision regret was mild [143] for both groups which likely reflects the effective medical management of croup and limited repercussions of the disease.

The parents who received the story booklets reported resolution of croup symptoms earlier compared to those receiving the standard information sheets. There are at least two possible explanations for this observation as well. First, the children's symptoms may have resolved more quickly due to how the parents managed the child at home following the ED visit. Alternatively, the parents' perception of the child's symptoms may have been affected by the intervention. For instance, if the parents who read the story booklets felt more reassured or more confident in their knowledge or ability to manage the condition, they may have been less bothered by ongoing coughing, difficulty breathing, or other symptoms. Since this outcome was self-reported by the parents, there is also the potential for bias, or exaggeration of the effect. Self-reported outcomes are particularly problematic when blinding is not possible. While we could not blind parents to the intervention they received, we attempted to blind them to the study hypothesis, the nature of the intervention, and the intervention that the other group received; thus, we expect that bias resulting from self-reporting was minimized. The significant results for this outcome may have been due to chance, therefore require substantiation in future research.

There are a number of potential reasons for the observed lack of effectiveness of the story booklets compared to the standard information sheet for the remaining outcomes. These include the following and are discussed below: the clinical context (i.e., clinical setting, nature of the illness); the outcomes selected and/or the measurement tools used; the timing of enrolment vis-à-vis the parents' needs; the relatively short duration of the healthcare encounter, and specifically the short

time between pre- and post-assessments for the primary outcome; the interactive role of the parent with their child throughout the healthcare encounter; the comparator (i.e., lack of important differences between the study and comparison interventions); the challenge of quantifying through existing tools the effects of this type of intervention; shortcomings with the intervention itself; and, characteristics of the target audience.

One of the main reasons for lack of effectiveness may be the clinical context in which we studied our hypothesis. While croup may cause a certain level of state anxiety when the child first exhibits symptoms, or exhibits the most intense symptoms, the medical treatment is highly effective with very rapid results, the condition is transient, and there are no known ongoing or long-term effects of either the condition or the treatment. Therefore, there may be a "ceiling" effect for this condition such that large differences would be unexpected regardless of the intervention. For instance, the majority (87%) of study participants were very or extremely satisfied with the treatment and information they received in the ED. It may be difficult to achieve a higher level of satisfaction and such a difference may not be clinically or practically important. This was found in a previous study comparing various formats of providing nutritional information (conversational, testimonial, didactic); the authors commented that "the ability to detect differences was limited by the generally positive ratings of the messages, which reduced variability."[147] Future research should help identify "when and how narratives can be most effectively used," e.g., which clinical scenarios (mild versus extreme disease, chronic versus acute conditions).[98] Further, narrative may be most useful in situations that are emotionally intense, where one is less able to focus on or process "complex didactic information."[98]

The "ceiling effect" or lack of potential for improvement was particularly relevant for the primary outcome. After much deliberation, the study team chose parental anxiety as the primary outcome. Based on anecdotal and empirical evidence, we believed that parents would have a higher than normal level of anxiety when managing a child with croup. Evidence has also shown a higher than normal level

of anxiety among parents when dealing with other illnesses or medical interventions involving their children. However, the baseline measures of anxiety observed in our study showed that parents were within "normal" levels of state anxiety based on norms for the outcome assessment instrument we used (i.e., the State Trait Anxiety Inventory, STAI-S) which is considered "a reliable, well-validated measure of acute situational anxiety."[11] Nevertheless, since the anxiety levels were already low at enrolment, there was little room to effect a change or a difference in change between groups. A similar observation was made in a study comparing statistical versus narrative messages to encourage mammography.[38] The participants found the narrative version more engaging, but there were no differences in outcomes (beliefs, attitudes, or intentions) between groups. One reason postulated was that "the participants were not for the most part particularly resistant to the message provided;"[98] hence, how the information was formatted and presented may not have had a discriminatory effect in terms of the outcomes assessed. In summary, if the end-users are already within the "normal range" of the outcome of interest, then one intervention may offer no advantage over another.

The low level of anxiety observed at baseline may have resulted from timing of enrolment and some of the practical limitations we faced when implementing the study. It would have been ideal to have provided the interventions outside of the ED setting and prior to the occurrence of the child's condition. This was in fact suggested to us during pilot testing of the tools: parents commented that this would be useful information to distribute through a public health unit prior to an episode of croup. However, it would be extremely challenging, time consuming, and costly to conduct a trial in that context in terms of numbers of participants and the extensive follow-up required. The alternative that we proposed was to identify patients as soon as possible after they arrived at the ED. This was also challenging for several reasons. First, we were unable to approach patients in the waiting room due to issues of confidentiality. Second, it was deemed unethical to approach potential participants when the child had not yet been examined, hence a

diagnosis of croup had not yet been confirmed. Third, at one site the triage nurses had standing orders to administer dexamethasone if croup was suspected. Therefore, most of the participants in the study had already been seen by the triage nurse (96%) and had treatment ordered (68%) prior to recruitment. The low levels of baseline anxiety suggest that these two mechanisms may have been sufficient to effect normal levels of parental anxiety if they were in fact high at the time of arrival to the ED.

Brunnquell outlined phases of reaction to trauma or emergency.[18] Parents may have already been past the first phase, described as the "panic" stage, by the time they were recruited into our trial. Once at the ED and seen by the triage nurse, they may have already moved onto the second phase of "protest and regression in which an individual's typical defences are used to maintain emotional equilibrium;"[18] hence, the intervention would have less potential for impact if parents were able to manage their emotions, particularly their anxiety, through their own defences. Further, a key identified source of concern and anxiety is unpredictability.[18] This may have been less of an issue at the point when parents were recruited into our trial as they had already been seen and often their children had already received treatment.

The duration of the healthcare encounter may have been too short to demonstrate changes in anxiety. This was observed in a study evaluating a 3-phase educational-behavioural intervention for children admitted to a pediatric intensive care unit (PICU) and their mothers which included reading and discussing a story about a young child who successfully copes with a stressful hospitalization.[119] Melnyk et al. found no differences in anxiety among the mothers during the hospitalization. The authors posit that the duration of hospitalization may have been too short to show changes in anxiety and mood state. The duration of hospitalization was an average of 7 days compared to our study in which few patients were admitted and the length of time in the ED was on the order of hours rather than days.

Melnyk et al. identified the loss of parental roles as a major source of stress. This may be more applicable to conditions requiring highly intensive medical interactions, such as in the PICU where Melnyk's study was conducted. In the ED context with croup, parents maintain the primary caregiver role for their child and the child is rarely removed from the parent for tests or interventions; therefore, the parents in this context may show lower levels of anxiety than in other medical scenarios where the child is removed from the parent for periods of time (e.g., surgery).

As another explanation for lack of significant findings in their study, Melnyk et al. suggested the fact that the control group received an intervention resulted in decreased anxiety and negative mood; therefore, differences between groups were not apparent. They suggested that comparison with a "pure" control group receiving standard care only may have demonstrated a greater effect of the test intervention. This effect was observed by Larkey et al. when evaluating storytelling versus a risk tool for colorectal cancer screening.[103] Since much of the content was identical for both interventions, this may have attenuated "observable effects on subtle mediating factors such as fear and risk perception, as well as the intention outcomes."[103] Conversely, in an evaluation of an interactive videodisc to reduce HIV/STD risk behaviours, Noell et al. found significant differences in all outcomes (beliefs, intentions and attitudes, self-efficacy); however, the intervention was compared against a waitlist control (i.e., nothing).[126] This may have been a factor in our study, since all participants received interventions which overlapped in terms of content thereby minimizing the relative impact of one intervention over the other.

The choice of outcomes in our study was based on factors that could be easily measured and quantified. If stories have an effect it may be on factors that are more challenging to measure. A priori we had a sense that the stories would provide greater overall comfort to the parents (or attend to their emotional reactions), although this construct was difficult to define and quantify. Therefore, with the intent to generate hypotheses, we chose a variety of outcomes that we felt

were related to this construct in different ways including anxiety, satisfaction, decision regret, and impact of the event. Many of these measures may be inadequate to evaluate whether the intervention "works." Petraglia also observed that the effects of narrative are difficult to measure and questioned what it means for a narrative intervention to "work."[132] He highlighted the chasm between the apparent power of narratives and current scientific understanding.

Another explanation for the results observed could be shortcomings with the intervention itself. There are many aspects of a story or narrative that can influence its impact and uptake. In the context of cancer prevention and control, Kreuter stresses that more research is needed to identify which attributes of the narrative (e.g., characters or messengers) enhance the likelihood of effecting the desired outcomes.[98] In the public health domain, there is a focus on the issue of "reception" in developing narratives to effect behaviour change.[132] Schank described a number of determinants of narrative impact.[138] First the end-user needs to be interested or care. The level of engagement will vary to the extent that the end-user understands the story and can relate to the story through their own personal experience. Second, the impact of a story is proportionate to the extent to which the end-user can identify with the main character or see themselves in that role.[138,147] The more details in the story, the greater the potential for the end-user to relate and identify based on their own memories—what Schank refers to as "triggering." Third, the timing of the story needs to coincide with the needs or desire of the end-user for the information in the stories. Schank asserts that "poor timing is one of the most common mistakes in educational environments."[138] Finally, Kreuter discussed "narrative quality," or whether the story is "told well."[98] This refers to how the different story elements are presented, including the order and context of each, and how well they match the objectives and preferences of the end-user. Larkey suggested that it may be important to measure potential mediating factors, such as "story appeal, identification and transportation by or engagement in the story" in order to understand why the intervention is or is

not effective.[103] We will attempt to do this through a qualitative analysis that was conducted alongside the trial.

A final explanation for the observed results is the target audience. While stories can enhance recall of information, Kreuter claims that the "advantages may be modest when the audience for such information is highly motivated and has education or experience with which to make sense of complex, didactic information."[98] In our sample, the education level was relatively high with the vast majority of parents having graduated from high school and over one half having some post-secondary education. Many narrative or story interventions have been developed and evaluated within populations of lower education and literacy levels, lower socio-economic status, and sometimes those with "a distrust of authorities."[59] These populations are often the most difficult to reach [59] and may not have other resources or access to other sources of comprehensible information. This underscores the need for the intervention to "match" the audience in terms of needs and levels of comprehension.[98] Our study population may have had access to other sources of information, such as the internet; in fact, 20% of the participants read information other than the study materials after discharge from the ED.

5.2 Future Research

Our experience conducting a trial in this topic area has lead to a number of recommendations for future research. First, researchers need to clearly identify the purpose of the stories prior to development and evaluation. In the context of cancer prevention, Kreuter identified "four distinct capabilities of narrative: overcoming resistance, facilitating information processing, providing surrogate social connections, and representing emotional and existential issues."[98] The purpose and timing of the intervention need to be matched to the needs of the end-user,[54] and these should ideally be identified through a systematic process. Often there are "incongruencies between what the patient wants and what health providers believe they want/need."[54] The identification of the end-user needs will

then directly inform the outcomes chosen to assess the effectiveness of the intervention. For example, if end-users want more information about a condition and its management, the focus of the stories and outcome assessment may be knowledge. If the end-users want a tool to facilitate decision-making, the choice of outcome may be decision regret. If the end-users want re-assurance, then some measure of anxiety may be appropriate. In some situations the end-user of the tool may be different from those commissioning the stories: for instance, health services administrators may employ stories to optimize healthcare and other resource utilization. The outcomes of most importance will also vary by the needs of the end-users, as well as the clinical context.

The outcomes selected for evaluation need to be assessed using validated, objective tools that are sensitive and specific to changes in the intended outcomes. McPherson commented on the outcomes assessed within the literature examining methods of providing information for cancer.[118] Many of the outcomes assessed were subjective, such as patient preferences, attitudes, uncertainty, and satisfaction. Often the measurement tools were designed by the investigators and tailored to the specific intervention under study. Subjective outcomes are more likely to lead to biased estimates of effect, particularly in research in this area where blinding is challenging. Appropriate methods need to be implemented to protect individuals in a study from knowing what intervention the participants receive. The use of cluster randomized trials may be particularly relevant, although these often require increased resources and present added logistical challenges. Many of the subjective outcomes are preferred as they can be assessed short-term, often immediately after the intervention is read by the patients. Research involving long-term and end-point outcomes (e.g., behaviour change) is required [115,126] in addition to process or intermediate outcomes (e.g., attitude, knowledge). While these longer-term outcomes (e.g., symptom management, health service utilization, patient coping) are often thought to be indirectly related to the intervention,[118] they are likely more important to the decision-makers that can influence whether the interventions are implemented in practice (e.g.,

physicians, other healthcare professionals, policy-makers in health services). Further, McPherson noted that "instruments of known reliability and validity" were more often used for these types of outcomes, thereby increasing the validity of the results stemming from their use.[118]

Another limitation of short-term assessments is that they may not allow sufficient time for the intervention to have an impact, particularly for "psychological indices."[118,119] Timing of providing the stories is a key factor identified by a number of researchers.[20,118] For example, if the information contained in the intervention is intended to influence the healthcare encounter (e.g., decision-making during the encounter), then participants may require the information prior to the healthcare encounter in order to allow sufficient time to review and absorb the information. Alternatively, if the information is intended to impact behaviour following the healthcare encounter, then provision of the information during the healthcare encounter may be appropriate. In the latter case, sufficient follow-up is required to examine whether there was in fact a change (for example in behaviour), as well as whether any change is maintained over time.

This topic area presents a unique challenge in that the development and pilot testing [147] of the stories and how they are packaged is a critical step. In many cases, there has been a disproportionate amount of effort and attention "into developing the narrative without understanding how it is received;"[132] this may stem from the assumption that stories are generally engaging.[74] In fact, Slater asserts that "success is unlikely without investment in formative research to develop effective characters and situations and to pretest the narratives."[147] There are a number of characteristics of the intervention that may influence its effectiveness. The primary consideration is the ability of the intervention to "transport" the reader and has been defined as the "integrative coding of attention, imagery, and feelings, focused on story events."[59,73] There are a number of factors that influence the ability of a story to transport the reader including: the quality of the story; readability and level of language; length and format; use of suspense and imagery; perceived realism and proximity to the reader; real-life versus

fictional accounts; ability of the recipient "to create vivid mental images;" and, the capacity of the story to create emotion on the part of the recipient, particularly empathy with the main character.[9,20,33,59,73,98,115,146] In order to maximize these factors, there needs to be a clear understanding of the end-user, including their values and experiences. These aspects will vary by culture; metaphor and symbolism within stories needs to be carefully crafted to match the beliefs and prior experiences of the end-users.[4,74] A further challenge arises when the end-user group is heterogeneous in its needs and preferences. One option is to create a single product – a "one size fits all approach" [97] – geared at different levels of understanding.[74] The other extreme is to tailor products to individual needs and personal characteristics. Tailoring of interventions has met with some success but requires more investment in terms of development and more sophisticated technology (e.g., computer technology) that may not be widely accessible by the target audience.[126]

Another challenge is that there are numerous aspects of the interventions that can be varied and studied, such as the medium of delivery (e.g., booklets, video, computer), length, writing style, and presentation (e.g., illustrations, images, colours, shape, and size).[20] These should be driven by the preferences of the end-users. In developing these products, there also needs to be a balance between the anecdotal and pragmatic or statistical information provided. There is substantial research comparing anecdotal/narrative versus statistical evidence with varying results;[4,73] however, Hinyard advises that "rather than arguing the merits of each…, it seems more productive to consider for whom and under what circumstances each might be most effective and how and when they might be combined to achieve optimum effects."[73] The balance of statistical versus anecdotal information will be dependent on the target audience and outcomes. Research suggests that the story first needs to be plausible (i.e., believable) in order for the information to be accepted, whether that information is anecdotal or statistical.[59,146] There is also the challenge of striking a balance between the comprehensiveness of the information, character and plot development with

optimal length and story structure.[98,147] Each of these factors may vary by the audience, their needs, and the targeted outcomes.

The bulk of the literature on the use of narratives and stories in healthcare is anecdotal or qualitative, with an important lack of randomized controlled trials.[118] Further, the intent of the interventions under study and outcomes assessed are varied which limits comparisons and overall conclusions for this type of intervention. A combination of quantitative and qualitative methods will be an asset in this area in order to evaluate effectiveness and to understand the mechanism through which the intervention acts, respectively.[118] There is also a need to understand the connection between exposure to information versus uptake and application of the information (e.g., for decision making or behaviour change).[4]

Finally, careful consideration is needed for the study comparison. Significant differences in this literature have more often been found when the intervention was compared against standard care or waitlist control, whereas fewer differences have been observed when compared to another active intervention.[126] To evaluate effectiveness, new interventions should be compared against the existing standard of care. Evaluations of specific aspects of the interventions are more challenging, and may be driven by user preferences identified through qualitative methods.

5.3 Strengths and Limitations

While formative research is essential, this study has gone beyond and studied the interventions in the context in which they will be used. This study represents an important step in terms of evaluating a non-medical intervention within the accepted biomedical model of investigation. We developed our intervention through an iterative process which involved pilot testing among healthcare professionals for content validity and focus groups of parents for appeal and readability. The intervention included three stories, each targeting a different severity of croup and with different main characters (both male and female) and situations.

We implemented a randomized controlled trial representing the highest level of research evidence. Further we followed accepted methods to avoid bias arising from inadequate allocation concealment. Blinding was a challenge due to the nature of the intervention. We blinded the participants to the study hypothesis and the interventions being compared. Where possible, we used validated tools to measure outcomes. While the results reported herein are primarily short-term, we did measure time to resolution of symptoms as well as anticipated anxiety should participants encounter a subsequent episode of croup. Further, we are currently collecting data one-year post-ED visit to measure long-term impact in terms of knowledge and resource use for subsequent episodes. We are also in the process of collecting qualitative data to gain insight into the mechanisms through which stories may or may not be effective.

One limitation is that our focus was on effectiveness in terms of benefits; we did not consider potential harms of the intervention as it was initially thought to pose no risks. However, during the development of the story booklets, we realized that there may be risks depending on how the readers choose to use or react to the information they are given. Our finding of significant decision regret among the story group leads to speculation as to whether they may choose not to go to the ED in the future when it may actually be necessary for the appropriate care of an ill child. Potential harms should be considered in future work.

Despite the scientific rigour of the present study, numerous questions remain for future work. Petraglia posed the question, "how does an intervention technique whose effects are so utterly and unapologetically subjective defend itself to administrators and funding agencies with biomedical expectations of scientific rigor?"[132] His answer is "perhaps, poorly" yet we believe of critical importance in order to obtain funding and encourage uptake of such interventions in the medical field. He further comments that "determining the benefits of a narrative intervention always will be a matter of piecing together an array of empirical evidence into a theoretically sound argument directed toward a particular audience."[132] This study adds data to the evidence base.

To date "there has not been a framework for organizing what is known (and not known) about how, when, and for what outcomes and audiences narrative health communication might be most effective."[98] There is an urgent need for this in order to advance research and knowledge in this area. The results and experiences gained through this trial will help towards this framework and understanding. There are two levels at which evidence should be collated. First, Mazor and Hinyard suggested that research be organized "according to the basic components of communication:" source (identifying with characters in story), message (fact versus fiction, first versus third person, more or less interaction, different narrative forms, dose), channel (print, TV, video, computer), and receiver (i.e., the target audience or end-user).[73,115] This will help elucidate the appropriate structure and mode of delivering stories. The second level is to develop a matrix in terms of the context in which stories have been examined, such as the care setting (acute, chronic, palliative, public health), types of conditions (acute, chronic, self-limiting), and target outcomes (e.g., knowledge, behaviour change, healthcare utilization).

The results obtained through this research represent an important advancement for knowledge translation in understanding whether storytelling is an effective means for transferring information to patients and their families. Further, the experiences gained through implementation of a trial in this topic area will serve to enhance the methodological rigour and relevance of future research. These results will inform subsequent steps including the development and evaluation of stories in other clinical areas and for specific cultural groups, as well as the development of other story-based communication tools.

Chapter 6

Risk of Bias in Pediatric Trials

6.1 Context

During the design and conduct of the storytelling trial, a number of questions arose around methods to prevent bias in order to yield the most accurate estimate of an intervention's effect. Two specific items that presented a challenge due to the nature of the intervention was blinding of the participants and study personnel, and unit of randomization (individual versus cluster) to prevent or minimize contamination. During the conduct of the trial, a new tool was released by The Cochrane Collaboration to assess risk of bias in randomized trials.[71] We applied the risk of bias tool to a sample of pediatric trials that were presented at the annual scientific meetings of the Society for Pediatric Research between 1992 and 1995. The results provided information on application of the tool and a comparison with other methods of assessing the methodological quality of randomized trials. The results also provided insightful data on the methodological quality or risk of bias in pediatric trials. Overall, only 6 of 163 trials in the sample were rated as low risk of bias. Tables 6.1 and 6.2 provide detailed results of the risk of bias and methodological quality across the sample of trials for different domains (e.g., allocation concealment, blinding, etc). Figure 6.1 provides some evidence that trials at high or unclear risk of bias result in larger treatment effects compared to trials at low risk of bias. This work provided the basis for: a proposal for empirical work regarding the impact of risk of bias on effect estimates in pediatric trials (which was the focus of the PhD Candidacy Examination); further evaluation of the risk of bias tool (oral presentation at the 17th Cochrane Colloquium, Singapore, October 2009); and, comparison with more recently published pediatric trials (poster presentation at the 17th Cochrane Colloquium, Singapore, October 2009). This work will also inform the development of standards for the design, conduct, and reporting of trials in child health, an international initiative being undertaken by StaR Child Health (http://www.ifsrc.org/).

The study as described in this chapter was recently published in the British Medical Journal.[67] Authors and their contributions include: Lisa Hartling (study

conception and design, project coordination, risk of bias assessments, data interpretation, drafting manuscript); Maria Ospina (study conception and design, risk of bias assessments, data interpretation, drafting manuscript); Yuanyuan Liang (data analysis, drafting manuscript); Donna M. Dryden (risk of bias assessments, data interpretation, drafting manuscript); Nicola Hooton (risk of bias and quality assessments, drafting manuscript); Jennifer Seida (risk of bias assessments; drafting manuscript); Terry P. Klassen (study design, data interpretation, critical review of manuscript). The authors acknowledge other contributions in the publication.

6.2 Background

Systematic reviews are considered the most comprehensive way for judging whether a treatment "does more good than harm."[3] The methodological quality of studies included in a systematic review can have a substantial impact on estimates of treatment effect, which may affect the validity of the conclusions of a review.[161] Careful consideration and appraisal of the methodological characteristics of the primary studies is an essential feature of systematic reviews. It helps to identify areas of strength and weakness in the existing evidence [128] and to formulate recommendations to improve the conduct and value of future research.

The terms "quality," "validity," and "bias" [71] have been used interchangeably in the systematic review literature to describe methodological conditions that are associated with the validity of study results. Traditionally, quality assessment in systematic reviews has primarily involved the appraisal of internal validity, that is, how well the study was designed and executed to prevent systematic errors or bias. Bias can result from flaws in the design, conduct, analysis, interpretation, or reporting of a study. In randomized controlled trials, bias has been classified into four general categories: selection, performance, detection, and attrition.[84]

Control of bias in randomized controlled trials is necessary to reduce the risk of making incorrect conclusions about treatment effects.[57] A number of empirical

studies have documented how the lack of adequate randomization, concealment of allocation, double-blinding, and differential losses to follow-up or dropouts per treatment group may affect the observed treatment effects.[25,84,92,120,139,140] Several "meta-epidemiological" studies have examined the effect of certain methodological characteristics and biases of individual randomized controlled trials on the pooled estimates of meta-analyses.[10,25,84,92,120,139,140] While the findings have been inconsistent across individual studies, there is evidence that inadequate or unclear allocation concealment and lack of double-blinding lead to exaggerated estimates of treatment effects.

The approach to quality assessment in systematic reviews is inconsistent and often debated.[84] The uncertainty regarding how quality measures are associated with estimates of treatment effect and the absence of a gold standard to assess the validity of randomized controlled trials [88] have resulted in the development of a large number of quality assessment tools.[3,121] Only 12% of the available scales and checklists to assess the methodological quality of randomized controlled trials have been empirically evaluated.[121] Further, these tools [85,88] often contain elements related to reporting (e.g., was the study population described) and design (e.g., was a sample size calculation performed) that are not related to bias.[71]

In February 2008, The Cochrane Collaboration introduced a new Risk of Bias tool to assess the internal validity of randomized controlled trials.[71] The tool was developed to address some of the shortcomings of existing quality assessment instruments. Specifically the tool was developed to assess the degree to which the results of a study "should be believed."[71] The choice of components for inclusion in the tool was based on empirical evidence demonstrating their association with effect estimates.[27,139,140] Furthermore, the developers aimed to distinguish between actual methods of conducting the randomized controlled trials rather than reporting.

The Risk of Bias tool is based on six domains: sequence generation, allocation concealment, blinding, incomplete outcome data, selective outcome reporting, and

"other sources of bias." Critical assessments (i.e., high, low, unclear risk of bias) are made separately for each domain. A final overall assessment within or across studies is based on the responses to individual domains. The assessments are to be made based on the trial report as well as additional documents, such as the study protocol. Those conducting the assessments are required to record the reasons for their decisions. In this way, the rationale for any judgments is documented and transparent.

Although the use of the Risk of Bias tool has been recommended for systematic reviews conducted within The Cochrane Collaboration, it has not been formally validated and it is unknown how the tool compares to other approaches currently available to assess the validity of a study. The objectives of this study were to evaluate: 1) the inter-rater agreement of the Risk of Bias tool; 2) the concurrent validity of the Risk of Bias tool compared to the Jadad scale [81] and Schulz [139] approach to allocation concealment; and 3) the relationship between overall risk of bias as assessed by the Risk of Bias tool and study effect estimates. Further, we planned to compare the time required to apply the Risk of Bias tool versus the Jadad scale and Schulz allocation concealment.

6.3 Methods

This cross-sectional analytical study was conducted on a convenience sample of 163 full manuscripts of randomized controlled trials in child health; these manuscripts resulted from abstracts that were presented at the annual scientific meetings of the Society for Pediatric Research between 1992 and 1995. The trials were part of a previously published project examining publication bias.[95] Their methodological quality had been previously assessed using the Jadad scale and Schulz allocation concealment.[81,139] Likewise, effect estimates for the primary outcome in each trial had been extracted.

A random sample of 80 randomized controlled trials were selected and evaluated independently by two reviewers (LH, MO) to assess the time to complete the Risk of Bias tool. This preliminary evaluation also helped to develop some guidelines

for application of the tool to the entire sample of trials. A single reviewer (NH) recorded the time required to apply the Jadad scale and Schulz allocation concealment to the same sample of 80 trials. Two reviewers (LH, MO, DD, NH, or JS) independently applied the Risk of Bias tool on the remaining trials following pilot assessment and discussion of five trials among the group of reviewers.

The primary outcome selected for each trial was used for those items in the Risk of Bias tool that require an outcome-focused evaluation (i.e., blinding and incomplete outcome data). We applied the tool based on instructions in the *Cochrane Handbook* [71] and consulted one of the developers of the tool (Dr. David Moher) for clarification as needed. For the "other sources of bias" domain, we assessed potential bias due to baseline differences, inappropriate influence of the study sponsor, and early stopping for benefit. For cross-over designs, we also considered whether such a design was appropriate and whether the wash-out period was sufficient.[71] Overall risk assessments (high, unclear, low) were based on the approach presented in the *Cochrane Handbook.*[71]

We assessed inter-rater agreement for each domain of the Risk of Bias tool and for the final overall assessment using weighted Kappa (k).[34,102] We categorized agreement as: poor (0.00), slight (0.01-0.20), fair (0.21-0.40), moderate (0.41-0.60), substantial (0.61-0.8), or almost perfect (0.81-1.00).[102] Correlations between domains of the Risk of Bias tool, the Jadad scale and Schulz allocation concealment were calculated using Kendall's tau statistic to assess the concurrent validity of the Risk of Bias tool. We also assessed the degree of correlation for the overall Risk of Bias assessment versus Jadad overall score; overall Risk of Bias assessment versus Schulz allocation concealment; and, high/low risk as assessed by Risk of Bias versus low/high quality as assessed by the Jadad overall score (i.e., score <3 versus ≥ 3 [120,122]). Time to apply the Risk of Bias tool and time to apply Schulz allocation concealment and the Jadad scale were compared using the paired t-test.

Effect sizes were calculated using Cohen's d for continuous outcomes; for dichotomous outcomes, the odds ratios were converted into effect sizes using a method devised by Hasselblad and Hedges.[68] The effect sizes were combined under DerSimonian and Laird random effects model.[44] Statistical heterogeneity was quantified using the I-squared (I^2) statistic.[72,141] Meta-regression was used to evaluate the effect of risk of bias on the effect size while controlling for possible study-level confounders including study type (efficacy versus equivalence), study design (crossover, factorial, or parallel), and outcome type (binary versus continuous, objective versus subjective). Studies were defined as efficacy versus equivalence based on "authors' statements with respect to the primary hypothesis."[95] To determine outcome type (objective versus subjective), two reviewers (LH, SC) classified the outcomes according to published guidelines [166] and reached consensus through discussion. Analyses were conducted using the Statistical Analysis System version 9.1 (SAS Institute, Carey, NC), the Statistical Package for the Social Sciences version 11.5 (SPSS, Inc., Chicago, IL), SPlus version 8.0 (Insightful Corporation, Seattle, WA), and Intercooled Stata version 7.0 (Stata Corporation, College Station, TX).

6.4 Results

6.4.1 Inter-rater agreement: The detailed risk of bias assessments by components are presented in Table 6.1. Similar summary information for Jadad and Schulz allocation concealment are in Table 6.2. Inter-rater agreement for the individual domains of the Risk of Bias tool ranged from slight (k=0.13 for selective reporting) to substantial (k=0.74 for sequence generation) (Table 6.1). Discrepancies were largely driven by reliance on reporting versus judgment regarding risk of bias. Hence, domains that involved a greater degree of subjective judgment regarding the potential risk of bias (e.g. blinding) tended to have poorer inter-rater agreement than domains that were more objective (e.g. sequence generation). For example, the same level of blinding in a study could yield more or less biased results for different outcomes: a hard end-point (e.g. mortality) may always be at low risk of bias regardless of the extent of blinding; for a subjective

outcome (e.g. quality of life) bias may be more likely if blinding of patients and caregivers was inadequate. Table 6.3 itemizes some of the sources of discrepancies and recommendations on how these might be addressed.

Table 6.1 Inter-rater agreement of the Risk of Bias tool

Domain	Risk of Bias Assessments			Weighted Kappa (95% CI)
	High	Unclear	Low	
Sequence generation	4	107	52	0.74 (0.64 to 0.85)
Allocation concealment	5	105	53	0.50 (0.36 to 0.63)
Blinding	16	49	98	0.35 (0.22 to 0.47)
Incomplete data	85	52	86	0.32 (0.19 to 0.45)
Selective reporting	16	19	128	0.13 (-0.05 to 0.31)
Other sources of bias	15	85	63	0.31 (0.17 to 0.44)
Overall risk of bias	61	96	6	0.27 (0.13 to 0.41)

Table 6.2 Jadad and Schulz Allocation Concealment Assessments

Domain	n (N=163)
Jadad	
Described as random	163
Randomization method	
Appropriate	47
Inappropriate	4
Described as double-blind	53
Double-blind method	
Appropriate	17
Inappropriate	2
Withdrawals/drop-outs described	57
Overall score	
0	4
1	54
2	56
3	32
4	16
5	1
Schulz allocation concealment	
Adequate	59
Unclear	100
Inadequate	4

Table 6.3 Sources of discrepancies and recommendations for selected domains of the Risk of Bias tool

Domain	Source of discrepancy	Recommendation
Blinding	Previous tools judge this domain based on reporting. In the Risk of Bias tool, reviewers make a judgment regarding the potential risk of bias associated with the level of blinding depending on the nature of the outcome.	• Identify outcomes (or groups of outcomes) to be assessed by this domain *a priori* • Develop guides for the interpretation and application of this domain based on the nature of the intervention and the outcomes chosen for the review
Incomplete data	Previous tools judge this domain largely on reporting. In the Risk of Bias tool, reviewers make a judgment regarding the extent of withdrawals, the reasons, and whether these two factors are likely to yield biased results.	• Identify outcomes (or groups of outcomes) to be assessed by this domain *a priori* • Develop guides for the interpretation and application of several factors: the proportion of withdrawals/drop-outs from the overall sample; the reasons for withdrawals/drop-outs; and whether the reasons and extent of withdrawals/drop-outs were different across study groups
Selective reporting	Ideally, one would compare the outcomes planned for a study (i.e., in the study protocol) with those that were analyzed and reported. The search and identification of study protocols may not be fruitful or feasible.	• In the absence of protocols or resources to locate protocols for each included trial, compare the outcomes described in the methods section to those reported in the results • Studies that report very few

		outcomes may also be at risk of selective reporting bias. *A priori*, identify the key outcomes that should be reported for the particular intervention and patient population.
Other sources of bias	Some of these include early stopping, baseline imbalance, differential diagnostic activity, contamination; some are based on trial design (e.g., cross-over, cluster, factorial). These items will vary according to the context and studies relevant to a given systematic review.	• Reviewers should decide *a priori* which 'other sources of bias' will be assessed and develop guides for interpretation • Consideration should always be given for: whether there were differences across groups in important variables at baseline; whether the authors declared their source of funding; and, whether a trial was stopped early for benefit

6.4.2 Time for Risk of Bias versus quality assessment: The mean total time to complete the Risk of Bias tool by two reviewers (including consensus) for a single outcome was 20.7 minutes (SD 7.6; range 11 to 58 minutes). Based on a sample of 80 trials, the mean time to complete the Risk of Bias tool by a single reviewer was 8.8 minutes (SD 2.2) compared to: 0.5 minutes for Schulz allocation concealment (SD 0.3; $p<0.001$); 1.5 minutes for the Jadad scale (SD 0.7; $p<0.001$); and, 2.0 minutes for Schulz allocation concealment and the Jadad scale combined (SD 0.8; $p<0.001$).

6.4.3 Concurrent validity of Risk of Bias tool: A high degree of correlation was found between the following domains: Risk of Bias sequence generation versus Jadad randomization; Risk of Bias allocation concealment versus Schulz allocation concealment; and, Risk of Bias blinding versus Jadad double-blinding (Table 6.4). Correlation was low for the following comparisons: Risk of Bias incomplete outcome data domain and the Jadad withdrawal item; Risk of Bias overall risk and total Jadad score; and, Risk of Bias overall risk and Schulz allocation concealment (Table 6.4).

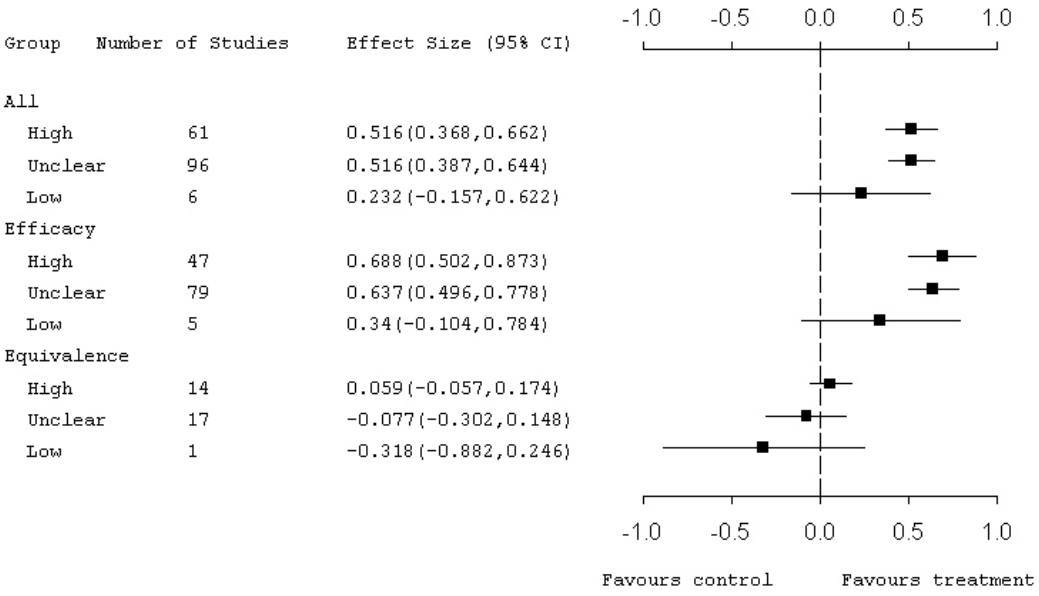
Table 6.4 Correlation between domains and overall risk as assessed by Risk of Bias versus Jadad Scores and Schulz Allocation Concealment

Comparison	Kendall's Tau
Comparison of Domains	
RoB sequence generation (yes/no/unclear) versus Jadad randomization (bonus/deduction)	0.788
RoB allocation concealment (yes/no/unclear) versus Schulz allocation concealment (adequate/inadequate/unclear)	0.729
RoB blinding (yes/no/unclear) versus Jadad double-blinding (bonus/deduction)	0.219
Incomplete outcome data (yes/no/unclear) versus Jadad withdrawals	-0.09
Comparison of overall risk or 'quality'	
RoB overall risk (high/unclear/low) versus Jadad (0-5)	0.059
RoB overall risk (high or unclear/low) versus Jadad (0-2/3-5)	0.085
RoB overall risk (high/unclear/low) versus Schulz allocation concealment (adequate/inadequate/unclear)	0.138

RoB=risk of bias

6.4.4 Relationship between risk of bias and magnitude of effect estimates: As shown in Figure 6.1, effect estimates were larger for studies assessed as having high or unclear risk of bias (high: n=61, effect size=0.52, 95%CI:0.37 to 0.66; unclear: n=96, effect size=0.52, 95%CI:0.39 to 0.64) versus those with low risk of bias (n=6, effect size=0.23, 95%CI:-0.16 to 0.62). We controlled for a number of potential confounders through meta-regression. The only variable that was statistically significant was study type (i.e., efficacy versus equivalence). The trend for efficacy studies was similar to all studies combined, where studies with high and unclear risk of bias had larger effect sizes than those with low risk of bias (high: n=47, effect size=0.69, 95%CI:0.50 to 0.87; unclear: n=79, effect size=0.64, 95%CI:0.50 to 0.78; low: n=5, effect size=0.34, 95%CI:-0.10 to 0.78). A reverse pattern was observed for equivalence studies, where those with high or unclear risk of bias were closer to the null compared to low risk studies (high: n=14, effect size=0.06, 95%CI:-0.06 to 0.17; unclear: n=17, effect size=-0.08, 95%CI:-0.30 to 0.15; low: n=1, effect size=-0.32, 95%CI:-0.88 to 0.25).

Figure 6.1 Effect size estimates according to risk of bias



6.5 Discussion

6.5.1 Principal Findings: We applied the Cochrane Risk of Bias tool to a sample of 163 pediatric randomized controlled trials. Despite guidance from the *Cochrane Handbook* on how to apply the Risk of Bias tool, the overall inter-rater agreement was fair. Our results stemmed from application of the tool by reviewers working in the same institution and review team. One might expect more variability across different research groups. This highlights the need for clear and detailed instructions to optimize reliability.

Much of the disagreement arose from items requiring judgment regarding the potential risk of bias given the methods or approaches described in a study. This underscores the need to establish clear guidelines at the outset of a review and to conduct pilot testing with a sample of studies that are representative of the review question or clinical area. In future research, we will examine whether decision rules can reduce inter-rater variability.

We found that the ratings for many domains of the Risk of Bias tool were "unclear." This may reflect the nature of the domain or insufficient reporting of study methods and procedures. In some cases, the assessment of "unclear" resulted from poor reporting at the individual study level. While reporting may improve for more recent studies as journals and authors adopt the Consolidated Standards of Reporting Trials (CONSORT) guidelines,[123] systematic reviewers will continue to face issues arising from poor reporting when they include studies from the pre-CONSORT era.

On average, it took experienced reviewers less than 10 minutes to independently apply the tool for a single, pre-determined outcome. The time required to complete the assessments may decrease with increased familiarity and use of the tool. However, more time will be required to apply the Risk of Bias tool in the context of a full systematic review, as assessments should be made for all main outcomes or classes of outcomes.[71] Furthermore, the *Cochrane Handbook*

recommends that study protocols are sought to inform or verify judgments.[71] This would further increase the time required to complete the Risk of Bias assessment.

There was a significant correlation between Risk of Bias and Schulz/Jadad in some domains (sequence generation/randomization, allocation concealment, blinding) but not others (missing data, overall scores). Higher correlations were obtained in domains that were most similar among the different tools. For example, the Jadad item evaluating whether the randomization sequence was adequately generated is similar to the sequence generation domain of the Risk of Bias tool. The lack of correlation for the missing data domain appears to be due to the emphasis on reporting in the Jadad instrument versus conduct in the Risk of Bias tool (i.e. how missing data were handled).

The lack of a significant correlation between the overall Risk of Bias and Jadad, and the Risk of Bias and Schulz allocation concealment, may reflect the different dimensions evaluated by the instruments. The Risk of Bias measures several domains that contribute to the overall assessment of risk of bias, including allocation concealment, and also incorporates selective outcome reporting and "other sources" of bias, domains that are not assessed by the Jadad scale. The lack of correlation could also be explained by the difference in how assessments are made; that is, the reliance on reporting for Jadad and Schulz allocation concealment versus the risk for biased results given the methods that were employed. The lack of correlation suggests that the different tools are measuring different constructs; hence, the Risk of Bias tool may be more appropriate for assessing a trial's internal validity.

A number of studies have provided empirical evidence demonstrating that trials with methodological flaws may overestimate treatment effects. This has been observed for allocation concealment,[45,92,128,139] sequence generation,[5,139] double-blinding,[139] handling of missing data,[133,157] and selective reporting of outcomes.[27-29] This study is the first to evaluate the Risk of Bias tool and to demonstrate its ability to differentiate between trials that may have overestimated treatment effects. Our results show that studies assessed as high or unclear risk of bias have

larger effect estimates than studies with low risk of bias. The pattern was consistent for efficacy studies, while the reverse pattern was observed for equivalence studies. These results should be considered cautiously given the small number of studies, particularly in the reference category. More rigorous statistical methods that minimize confounding due to intervention and disease are required to confirm these findings. Nevertheless, the results provide some preliminary validation of the Risk of Bias tool's usefulness to identify studies that may exaggerate treatment effects. This is particularly relevant to systematic reviewers as well as any practitioner who wants to assess the potential impact of an intervention.

6.5.2 Limitations: This study had several limitations. For efficiency, we used information that was generated as part of a previous study (i.e., effect size data; selection of a single, pre-specified outcome; previous Jadad and Schulz assessments).[95] As such, there was a time delay between application of the Jadad/Schulz and the Risk of Bias tools; moreover, the tools were applied by a different team of researchers. This may have contributed to some variability in the application and interpretation of these assessment tools and likely attenuated the observed correlations; however, it is likely that this more closely resembles the use of these tools in real settings. We applied the Risk of Bias tool to a single outcome, which is not the recommended approach. This may have resulted in some studies being rated differently, in terms of overall risk of bias, than if we had considered all of the main/important outcomes. While we found significant differences in effect sizes comparing high or unclear versus low risk of bias, these were based on small numbers of low risk studies (n=6 in total) and the confidence interval for the low risk studies was wide. Assessing a more recent, post-CONSORT sample of studies may increase the number of low risk studies and may provide a more certain estimate of the impact of risk of bias on effect size. Further, the studies in our sample were published prior to release of the CONSORT statement, which may have resulted in more "unclear" assessments than may be the case for more recently published studies. The sample of trials was heterogeneous in terms of outcomes, interventions, and diseases; this differs from

the hallmark meta-epidemiological studies in this area that have evaluated the relationship between methodological characteristics and effect estimates.[58,152] We used effect sizes to standardize the measures of effect so that we could look at general patterns across studies with different risks of bias. Finally, the sample included only pediatric trials; hence, the results may not be generalizable to other areas of health care.

6.5.3 Conclusions: We found substantial variation in agreement across domains of the Risk of Bias tool. Generally the items with poor inter-rater agreement were those that required substantial judgment regarding the potential for the study methods to yield biased results. There was low correlation between overall assessments using the Risk of Bias tool compared to two commonly used tools (Jadad and Schulz allocation concealment). Overall risk as assessed by the Risk of Bias tool differentiated effect estimates with more conservative estimates for low risk studies. Careful training and clear guidelines are required when applying the tool.

Chapter 7

Conclusions

The work contributing to this dissertation involved three main components: development and pilot testing of story booklets; a two-site randomized controlled trial to evaluate the effectiveness of the story booklets; and, an examination of risk of bias in a sample of pediatric trials. The results provide evidence for the effectiveness of one intervention and direction for future research to ensure valid results.

We followed a systematic process to develop and test story booklets for parents attending the ED with a child with croup. Our testing provided rich feedback and allowed us to shape our products to ensure accuracy, credibility, and relevance to the end-user. Our experience highlights many considerations for future development work in this area, including clear identification of the purpose and goals of the end-product at the outset and involvement of the end-user group throughout to identify needs and preferences. Moreover, our results are informative more broadly for the development of patient education materials and tools to communicate with patients. Whether stories are effective in practice needs to be assessed through rigorous, research methods. Mixed methods approaches that combine quantitative and qualitative data may be most useful in this context to measure effectiveness and explore underlying mechanisms, respectively.

We conducted a randomized controlled trial comparing story booklets versus standard information sheets for parents of children attending the ED with croup. There was no significant difference in the primary outcome of change in parental anxiety between the time of enrolment to discharge from the ED. The story group showed significantly greater decision regret and quicker time to resolution of symptoms; however, the clinical or practical significance of these findings is unknown and further research is required to substantiate these findings. No differences were observed for the remaining outcomes. There are a number of potential reasons for the lack of significant findings including choice of outcome, timing of outcome assessment, and disconnect between the nature of the intervention and the needs of the target audience.

This study adds to a growing evidence base for the use of stories to communicate with healthcare consumers. This is one of few randomized controlled trials to examine the use of stories and serves as a model for future research in this area. An examination of the risk of bias in a sample of pediatric trials demonstrates that there is substantial room for improvement in the design, conduct and reporting of research related to child health.

The lessons learned from this study in terms of narrative development, outcome selection, and risk of bias will provide solid direction for future research. These results provide critical information regarding the use of stories in the emergency department setting for an acute and self-limiting condition. This contributes to an evidence matrix identifying when, where, and for whom stories may be most effective.

Despite the limited significant results found in this study, Schank asserts that "stories will always be an integral part of our lives, for entertainment, communication, teaching, and learning." (Schank 2002) We need to understand and harness that power to effect change in the health care setting for improved health outcomes, quality of care, and resource utilization.

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APPENDICES

APPENDIX A. Search strategies for relevant background material, including systematic reviews and other trials

Medline
Searched January 30, 2006
---
1 exp Narration/
2 exp anecdotes/
3 exp medicine in literature/ or exp mythology/
4 storytell$.mp.
5 or/1-4
6 exp mental health/ or exp cognition/ or exp intention/ or exp learning/ or exp "mind-body relations (metaphysics)"/ or exp thinking/
7 5 and 6
8 exp Teaching/
9 7 and 8
10 exp Patient Education/
11 7 and 10
12 5 and 10
13 12 not 11 (60)
14 storytell$.ti. (114)
15 or/9,13,14

Embase
Searched January 30, 2006
---
1. exp Literature/
2. "knowledge uptake".mp.
3. consumer$.mp.
4. storytelling.mp.
5. or/1-4
6. vignette$.mp.
7. exp INFORMATION/
8. 6 and 7
9. exp Patient Information/
10. 6 and 9
12. narrative.mp.
13. 6 and 12
14. 9 and 12
15. or/5,8,10,13,14

Eric
Searched January 16, 2006
---
1 exp PARENTS/
2 story telling/

3 exp Teaching Methods/
4 or/1-3
5 exp Communication Research/
6 2 and 5
7 exp learning/
8 2 and 7
9 8 and 3
10 exp adult learning/
11 2 and 10
12 exp NARRATION/
13 exp Learning Processes/
14 2 and 13
15 2 and 12
16 or/4,6,8,9,11,14,15
17 clinical nursing research.mp. or clinical research/
18 control group/
19 random$.mp.
20 ((singl$ or doubl$ or tripl$ or trebl$) adj10 (blind$ or mask$)).mp.
21 (cross?over or placebo$ or control$ or factorial or sham$).mp.
22 (therapy or treat$).mp.
23 ((clin$ or intervention$ or compar$ or experiment$ or preventive or therap$) adj10 (trial$ or study or studies)).mp.
24 exp Experiments/ or clinical research.mp.
25 (clin$ adj25 (trial$ or study or studies or design)).ti,ab.
26 ((singl$ or doubl$ or trebl$ or tripl$) adj25 (blind$ or mask$)).ti,ab.
27 RESEARCH DESIGN/
28 (Follow up adj5 (study or studies or design)).ti,ab.
29 Follow up Studies/
30 Cross Sectional Studies/
31 Comparative Study/
32 Comparative Analysis/
33 exp Probability/
34 ((Allocat$ or control$ or assign$ or treatment or compar$ or interven$ or experiment$) and (group or groups)).mp.
35 (group or groups).ti,ab.
36 ((control$ or prospectiv$ or retrospectiv$ or evaluation or outcome$ or volunteer$ or participant$ or compar$) and (trial$ or study or studies or design)).mp.
37 cohort$.ti,ab.
38 case-control$.ti,ab.
39 Cross sectional.ti,ab.
40 (observational adj5 (study or studies or design)).ti,ab.
41 Longitudinal.mp.
42 Retrospective.ti,ab.
43 Relative risk.ti,ab.
44 Odds ratio.ti,ab.

45 (case adj (comparison or referent)).ti,ab.
46 (Causation or causal$).ti,ab.
47 (Analytic adj (study or studies)).ti,ab.
48 exp Evaluation Research/
49 cohort analysis/
50 or/49-81
51 2 and 50
52 limit 51 to ((adult basic education or postsecondary education or two year colleges or higher education programs or graduate study) and ("adult, career, and vocational education" or higher education or junior colleges or "reading and communication skills" or "tests, measurement, and evaluation"))
54. or/16,52

CINAHL
Searched January 18, 2006
---
1 exp STORYTELLING/ed, ev [Education, Evaluation]
2 exp storytelling/
3 exp adult education/ or exp learning methods/ or exp teaching methods/ (23388)
4 2 and 3
5 limit 4 to (adult <19 to 44 years> or middle age <45 to 64 years> or aged <65 to 79 years> or "aged <80 and over>")

PsycINFO
Searched January 18, 2006
---
1 exp storytelling/ (1993)
2 exp adult education/ or exp learning methods/ or exp teaching methods/
3 1 and 2 (84)
4 narrative teaching.mp. (10)
5 exp client education/ or exp health education/ or exp health knowledge/ (12038)
6 1 and 5
7 storytell$.mp.
8 5 and 7
9 7 and (2 or 5)
10 limit 9 to (("treatment (high sensitivity)" or "reviews (high sensitivity)") and adulthood <18+ years>)
11 or/3,4,6,8,10

Global Health
Searched January 17, 2006

Search strategy:
storytelling.mp.

Dissertation Abstracts
Searched January 18, 2006

#1 (storytell* AND adult learning)
#2 (storytell* AND communicat*) AND (medic* OR health* or clinic* OR patient*)
#3 (storytell* AND (educat* OR learn* OR communicat*)) AND (medic* OR health* or clinic* OR patient*)
#4 #1 or #2 or #3

Scopus
Searched January 18, 2006

Search strategy:
TITLE-ABS-KEY-AUTH(storytelling AND (uptake OR communicat* OR learn*))

MLA Abstracts
Searched January 18, 2006

Storytell* or narrative*

Social Sciences Abstracts
IBSS (International Bibliography of the Social Sciences)
Linguistics and Language Behavior Abstracts:
Searched January 13 and February 7, 2006

Search strategy:
(DE "Storytelling") or (DE "Storytelling--Psychological aspects") or (DE "Storytelling--Psychological aspects. nnnn") or (DE "Storytelling--Psychological aspectsnnnn") or narrative* or stories* or story or storytelling

AND

(DE "Health Education") or (DE "Medical Education") or "health education" or (educat* and medical) or "communication tool*" or (health and information)

Web of Science
Searched February 7, 2006

TS=(narrative* or stories or story or storytelling)

AND

TS=health education or TS=communication tool

APPENDIX B. INFORMATION SHEETS AND CONSENT FORMS

UNIVERSITY OF ALBERTA

Department of Pediatrics
Faculty of Medicine & Dentistry

INFORMATION FORM
PART A (Randomized Trial)

Title of Project: Communication Tools for Parents of Children Presenting to the Emergency Department with Croup

Lead Clinical Investigators: Dr. Terry P. Klassen, Stollery Children's Hospital
Dr. David Johnson, Alberta Children's Hospital

Co-Investigators: Dr. Shannon Scott-Findlay, University of Alberta
Ms. Lisa Hartling, University of Alberta

We are asking you to be part of a research study. This study will try to help improve trips to the emergency department for parents of children with croup.

This hand out is one part of you giving informed consent. A copy of this sheet has been given to you to keep. It lets you know what the research is about and what we would like you to do. If you would like more information you should feel free to ask. Please take the time to read this carefully.

Why are we doing this study?

Parents are becoming more involved in the healthcare of their children. It is important that parents receive information to help them understand their child's illness. This can help them better understand the illness, how it can be treated, and what they can do at home.

What will happen?

Participating in this study will involve:

a) completing a questionnaire that will take about 15 minutes of your time shortly after you agree to participate in the study;
b) completing a second questionnaire that will take less than 10 minutes when your child is discharged home;
c) completing a short telephone interview that will take about 15 minutes 1 and 3 days after your hospital visit, and every other day until your child is better or until 9 days after your hospital visit, whichever one comes first;
d) being contacted by telephone again one year after your hospital visit to complete a short questionnaire that will take 5 to 10 minutes.

If you choose to be in this study, you will be placed in one of two study groups. All parents will receive information on their child's illness, but how the information is given will be different for the two study groups.

1

Your time in the emergency department may be 10 minutes longer if you agree to be in this study. The medical care that your child gets will be the same as the care they will get if you do not choose to be part of the study. The medical care will not change.

Benefits and Risks:

You may not benefit directly from being in the study, but you will be helping us understand the best way to give information to parents of children with croup. We anticipate no risk of harm from the research study. The care your child receives will not change.

Consent:

You can say yes or no to any or all of the study parts. Answering the study questions is your choice. You do not have to answer any questions you do not want to. You should not feel any pressure from Emergency if you do not want to join the study. Your child's care will not be affected in any way. We would like your permission for the researcher to look at your child's chart and write down the first three digits of your postal code, your child's age, the date and time that you arrived at and left Emergency, and whether your child went home from emergency or was admitted to hospital. We would also like to use your Alberta Healthcare number to look at your child's healthcare records in one year's time to see if you have had other hospital visits for croup.

Confidentiality:

- Your information will not be shared with anyone in Emergency.
- All information in this study will be kept for at least seven years in a secured area. It will not be destroyed. The information may be looked at again in the future to help us answer other study questions. If so, the ethics board will first review the study to make sure that the information is used ethically.
- Only the research team will see your information.
- Your name, your child's name, and any personal health information will not be attached to your information.
- Your name or your child's name will never be used in any presentations or publications of the study results.
- All information will be held private, except when professional codes of ethics or the law requires reporting (i.e. child abuse)

Your signature on the information sheet lets us know you understand the information about being part of this study and agree to participate. If you have any further questions concerning to this study, please contact:

Dr. Terry P. Klassen (780-407-7084) or
Lisa Hartling (Study Coordinator; 780-492-6124)

Should you have any questions regarding your rights as a participant, you may contact the **Capital Health Patient Concerns Office** at 780-407-1040.

UNIVERSITY OF ALBERTA
Department of Pediatrics
Faculty of Medicine & Dentistry

CONSENT FORM

PART A (Randomized Trial)

Title of Project: Communication Tools for Parents of Children Presenting to the Emergency Department with Croup

Lead Clinical Investigators:	Terry P. Klassen (Edmonton)	Phone Number: (780) 407-7084
	David W. Johnson (Calgary)	Phone Number: (403) 955-7507
Co-Investigator:	Shannon Scott-Findlay	Phone Number: (780) 492-5074
Study Coordinator:	Lisa Hartling	Phone Number(s): (780) 492-6124

	Yes	No
Do you understand that you have been asked to be in a research study?	☐	☐
Have you read and received a copy of the attached Information Sheet?	☐	☐
Do you understand the benefits and risks involved in taking part in this research study?	☐	☐
Have you had an opportunity to ask questions and discuss this study?	☐	☐
Do you understand that you are free to withdraw from the study at any time, without having to give a reason and without repercussions to you?	☐	☐
Has the issue of confidentiality been explained to you?	☐	☐
Do you understand who will have access to your records, including information from you and about you?	☐	☐

Who explained this study to you? _____

I agree to take part in this study: YES ☐ NO ☐

Signature of Research Subject _____

(Printed Name) _____

Date: _____

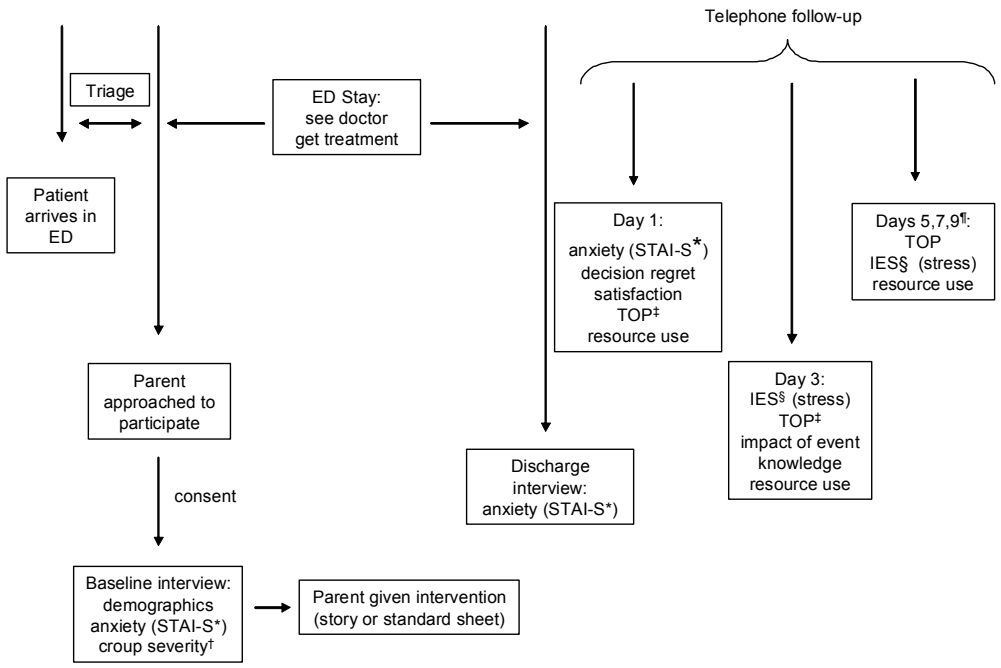
Signature of Witness _____

I believe that the person signing this form understands what is involved in the study and voluntarily agrees to participate.

Signature of Investigator or Designee _____ Date _____

THE INFORMATION SHEET MUST BE ATTACHED TO THIS CONSENT FORM AND A COPY GIVEN TO THE STUDY PARTICIPANT

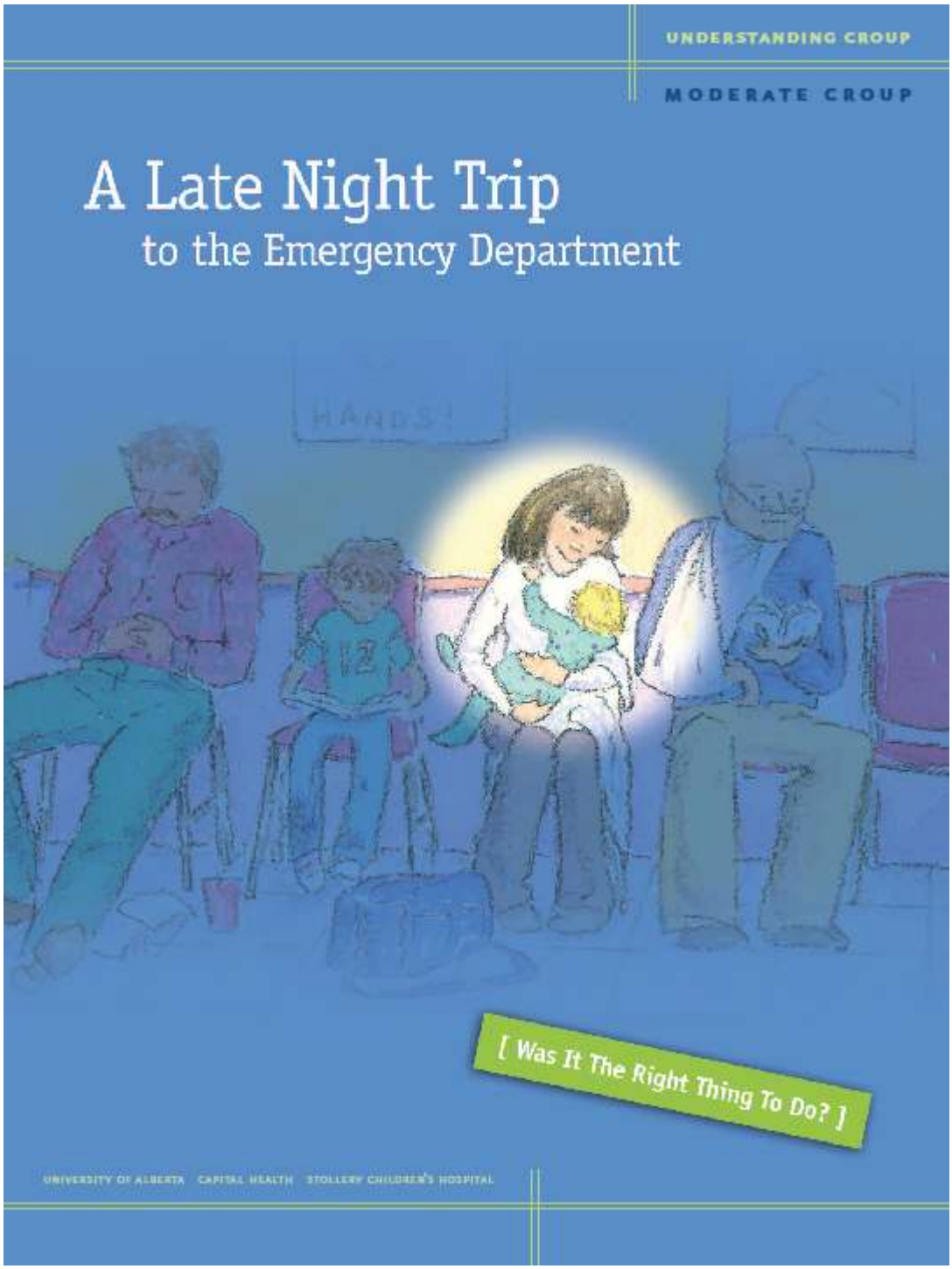
APPENDIX C. Flow diagram of patient recruitment and follow-up

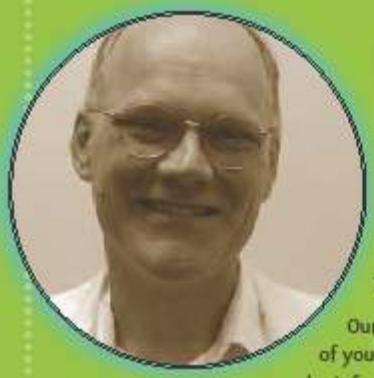


* State Trait Anxiety Inventory, (State Version: Form Y) (Spielberger CD. State-Trait Anxiety Inventory: a comprehensive bibliography. Palo Alto, California: Consulting Psychologists Press, 1984)
† Westley Croup Score (Westley CR, Cotton EK, Brooks JG. Nebulized racemic epinephrine by IPPB for the treatment of croup: a double-blind study. Am J Dis Child 1978; 132(5):484-7)
‡ Telephone Outpatient Score for Clinical Status (Johnson DW, Williamson J. Telephone out patient (TOP) score: the derivation of a telephone follow-up assessment tool for children with croup. Pediatr Res 2003; 53:185A)
§ Impact of Event Scale (Horowitz M, Wilner N, Alvarez W. Impact of Event Scale: a measure of subjective stress. Psychosom Med 1979; 41(3):209-18)
¶ Patients were followed up beyond day 3 if they still had symptoms on day 3; follow-up continued until symptoms resolved (i.e., TOP score = 0)

APPENDIX D. STORY BOOKLETS

BOOK ONE





[A Message from Dr. William Craig]

CROUP IS AN ILLNESS THAT AFFECTS YOUNG CHILDREN. It can come on quite suddenly and can cause severe coughing. In some cases, the child may have difficulty breathing. It is a stressful time for the parents of a young child.

Our first goal as health care professionals is to take care of your child. Another goal is to make sure that you have the information you need to understand your child's illness. Understanding the illness and how it is treated will ease the stress that you may have when your child is sick.

This booklet includes the story of a parent like you who had a child with croup. The story tells about one family's experience. The story also includes medical information on the illness and how it is treated.

A story told through the eyes of a parent is a novel approach to passing on information. Most of us, from the very young to the very old, enjoy a good story. Stories also help us remember details – more so than reading a textbook or a scientific report.

This booklet includes one family's story but not all cases of croup are the same. Some are very mild and can be treated at home. In other cases the child should be seen by a doctor or at the emergency department.

I hope that this story will help answer some of the questions you have about croup. I also hope that you will find interest in reading other parents' stories, and that you will find comfort in knowing that you are not alone in your experience with croup.

DR. WILLIAM CRAIG, MD
Director of Pediatric Emergency
Stollery Children's Hospital, Edmonton, AB

> Get Info <

For more information on croup and how it can be treated please visit the Alberta Medical Association website to find the Clinical Practice Guideline (CPG) on Croup: **www.albertadoctors.org**

Michael's barky cough startled Valerie from her sleep.

FOR THE LAST FEW NIGHTS, Valerie had not slept well as her two-year-old son was fighting a cold and had been up off and on with a fever and cough. Her first reaction was irritation – that she was going to have to spend another fitful night nursing her sick son. She desperately hoped the coughing would stop.

The lack of sleep was taking its toll. She was losing her patience with Michael and she knew she had been short with her customers at work. Couldn't they understand that she had more important things to deal with than their petty complaints? Her resentment towards her customers, and especially towards Michael, made her even more frustrated and guilty. Sometimes she just didn't feel that she was a very good mom.

More barky coughing roused her from her thoughts. This time the cough frightened her. As she ran into his room, Michael seemed to be fighting for breath.

In a panic, Valerie picked him up in her arms and ran to the phone. She grabbed the receiver, but it was already late and she wasn't sure who to call. She looked around the kitchen not knowing what to do. It was times like these when Valerie felt very alone as a single mom. Valerie was exhausted and at her wit's end – she couldn't handle one more sleepless night. She knew that she had to take Michael to the emergency department to get the help they both needed.

> Get Connected <

Nurse Telecare Line:

Capital Health Link in Edmonton 408-LINK

Calgary Health Link 943-LINK

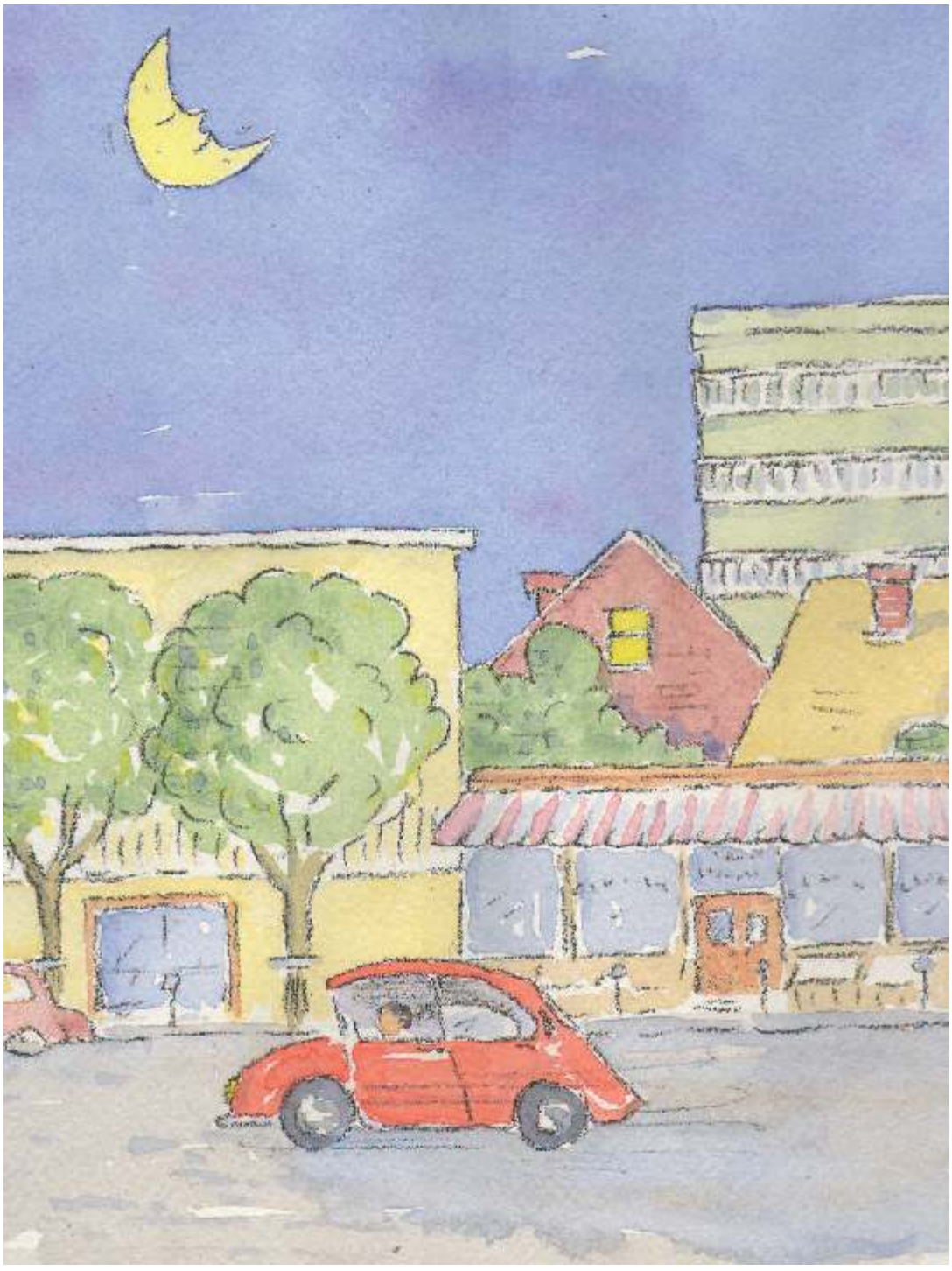
Outside the Calgary and Edmonton areas, toll free 1-866-408-LINK

Valerie wondered how she would get to the hospital. She didn't know whether the bus would be running at this late hour and she didn't have the money for a cab.

Though it was late, she reluctantly called her neighbour whose older daughter babysat when Valerie had to work late. Valerie explained her situation and they agreed to meet out front in ten minutes.

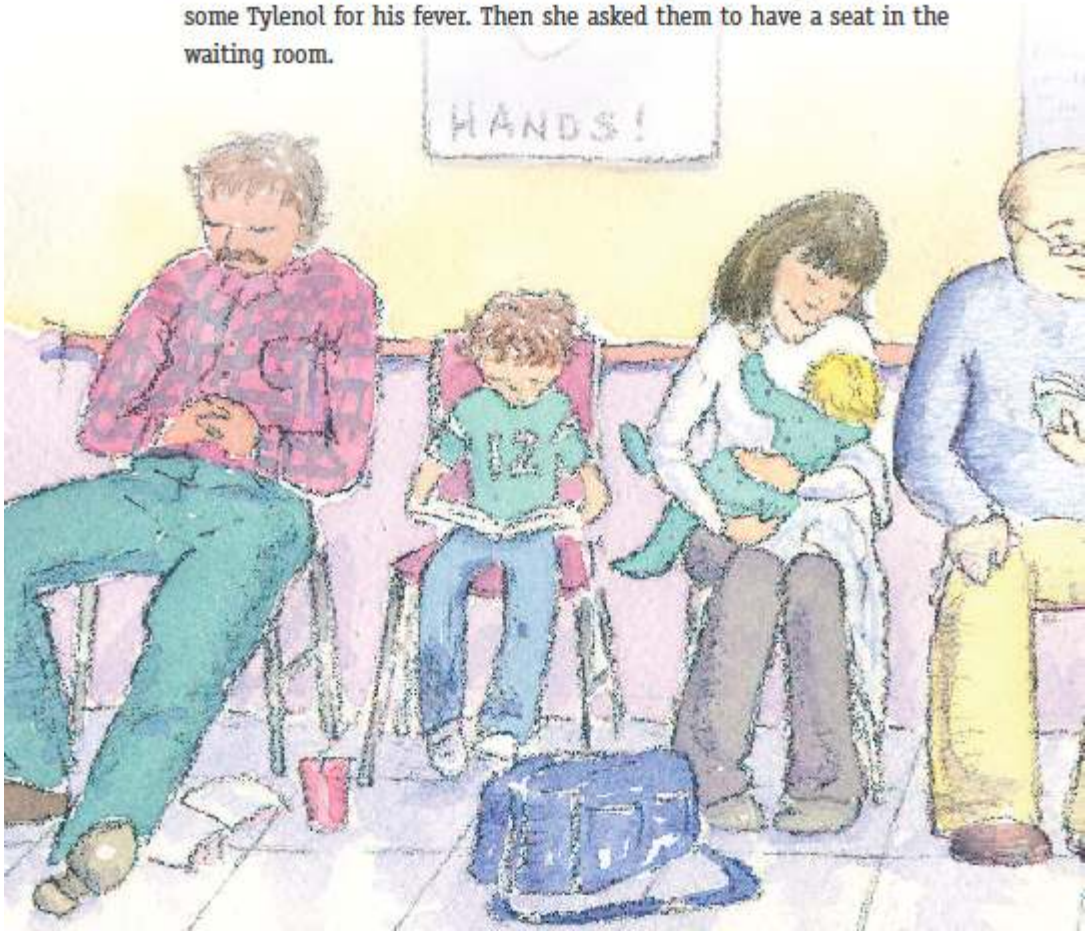
Valerie nervously watched Michael from the front seat of the car as they raced along the empty streets. Her neighbour rolled down the window to clear the windshield and within a few minutes, Michael's coughing got a little better. Though he was still breathing very loudly and his voice sounded scratchy, he seemed calmer and happier as he babbled to them from the back seat. She was surprised that he was so wide awake at this late hour, but figured it was because of the extra long nap he had taken that afternoon.

Take your child into cold air outdoors or sit by an open window or an open freezer door.



Just before midnight they walked through the emergency department doors. Within minutes of being in warmer air, Michael's breathing got worse.

He felt hot and his coughing was causing him obvious discomfort – his eyes were watering and his body trembled with each cough. Valerie sank down into the chair at the front desk and described to the nurse what had happened over the last few days. The nurse made notes on the chart and checked Michael. After talking to a doctor, she gave him some Tylenol for his fever. Then she asked them to have a seat in the waiting room.

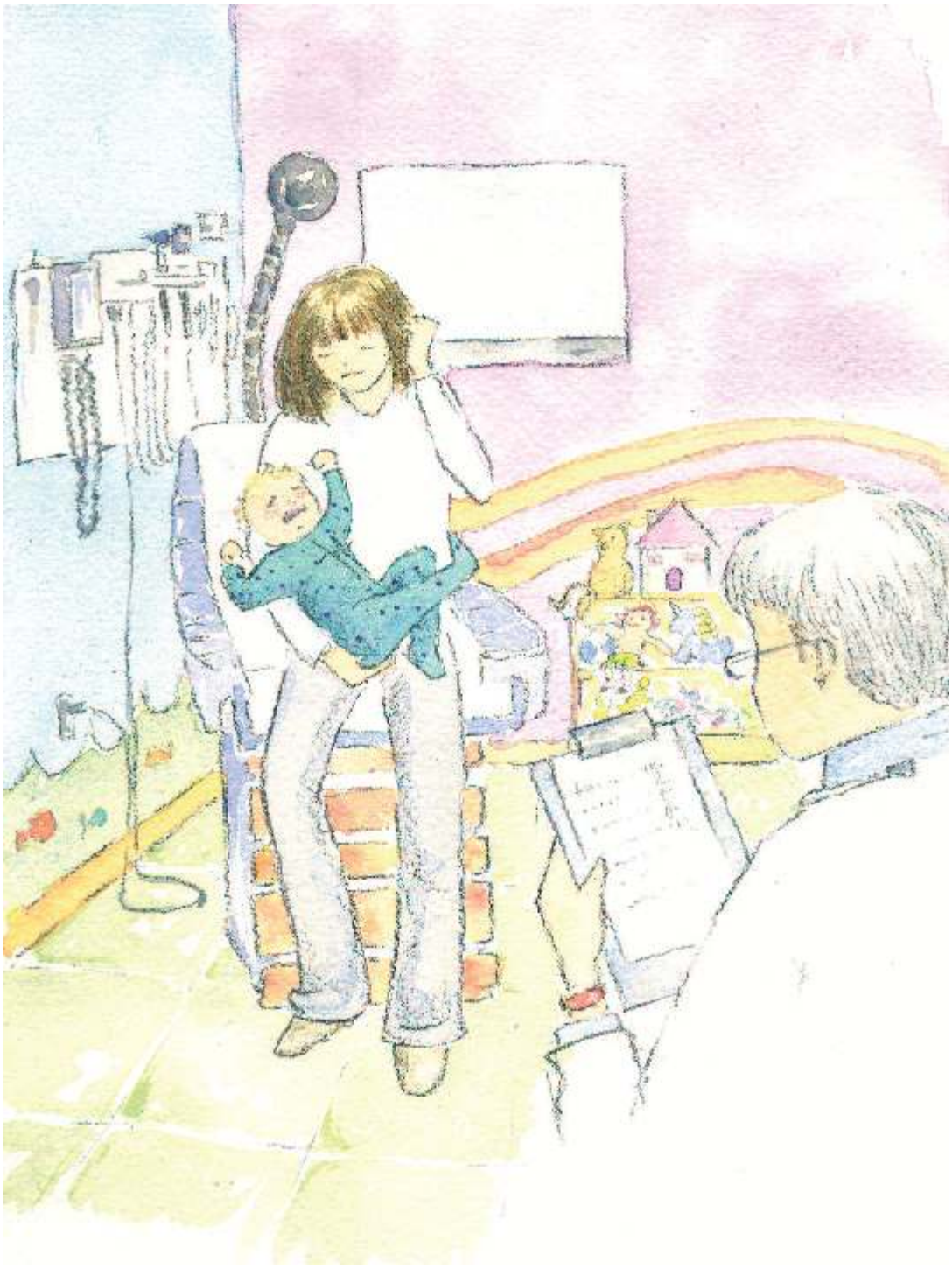


The emergency department was very busy and the wait seemed endless. Finally after three hours, Valerie and Michael were lead to an examination room. Michael was restless and hungry as he had refused to eat since late afternoon. Valerie gave him a bottle that she had packed. It did the trick. By the time the doctor arrived, Valerie doubted her decision to come to the hospital – Michael's breathing was back to normal and the other patients looked much sicker than her son.

The doctor quickly read Michael's chart before entering the examination room. The nurse had noted the classic symptoms of croup: several days of mild fever, a hoarse voice, and a seal-like barky cough that had started late this night.

"Another case of croup", the doctor thought to himself – he had seen several cases lately as it tends to cluster in the fall and winter months.

As he entered the room, the doctor noted that the mother looked tired and stressed. He realized that though this was another case of croup for him, it was an intense and worrisome event for the parent and child. The harsh barky cough that can rouse parents from their sleep often causes fear and alarm. The coughing is startling, especially when heard for the first time, and often sounds much worse than it actually is.



"What brings you here tonight?" the doctor asked in a friendly tone as he sat down by the desk.

Valerie looked up at him apologetically and declared, "I wasn't sure whether or not to come. He was coughing so much and seemed to be having such a hard time breathing at home. Now he already seems to be getting better."

The doctor reassured Valerie that she should feel good about her decision – after many years as a doctor, he knew the value of a mother's intuition.

Valerie explained to the doctor what had brought her to the emergency department. The doctor examined Michael and confirmed what he suspected – Michael was suffering from a moderate case of croup.

"Croup is an infection that is caused by a virus," the doctor explained. He added, "this can make the vocal cords, windpipe and voice box swell – it's this swelling that causes the hoarse voice and the barky cough".

"I recommend giving your son a steroid called **dexamethasone,**" the doctor explained. "The steroid will help with the swelling one or two hours after taking it."

Dexamethasone is a steroid that reduces swelling in the windpipe and voice box so the child can breathe easier and has a less harsh cough. It is a prescription medication prescribed by your doctor and available at most pharmacies.

"The nurse will bring the steroid for Michael. The medicine is sweet tasting and there are no serious side effects".

The doctor told her that they would watch Michael for a few hours and if all was well, Valerie and her son would be on their way.

Sure enough several hours later when the doctor returned, Valerie appeared much calmer and Michael was resting comfortably in her arms.

"What if the coughing gets worse again?" asked Valerie after the doctor told her they could go home.

"In most cases the cough is worse the first night and should be gone completely within 2 to 5 days," the doctor assured her. "You can give Michael something if he feels uncomfortable, like Tylenol, Tempra, Advil, or Motrin, but other medications such as cough syrups, decongestants, and antibiotics won't help with croup."

VALERIE GATHERED HER CHILD AND HER BELONGINGS AND PAUSED TO THANK THE DOCTOR AS SHE LEFT THE ROOM. She was so relieved that she could feel the stress drain from her body and she longed to be asleep in bed. The doctor watched them go and silently wished her a peaceful night, thinking back to the many sleepless nights he had experienced when his own children were young. ■

Signs of Croup

Croup symptoms most often happen in late evening and at night and start quite suddenly.

Croup often begins like a cold, but then fever, cough and difficulty breathing develop.

Stridor: when your child breathes in, you hear a harsh, vibrating sound. This gets worse when the child cries or coughs.

Barky cough: the child may sound like a dog or a seal

Hoarse voice

Cold symptoms: fever, runny nose, irritability, decreased appetite

Trouble breathing: the air passage is swollen, making it difficult for the child to breathe in and out.

Croup symptoms frequently improve on the way to medical care and may get better or worse depending on whether the child is calm or agitated.

Croup symptoms usually improve during the day, and often happen again the next night.

Most children are better within two days, but a small number of children have symptoms that continue for up to one week.

A Late Night Trip
to the Emergency Department

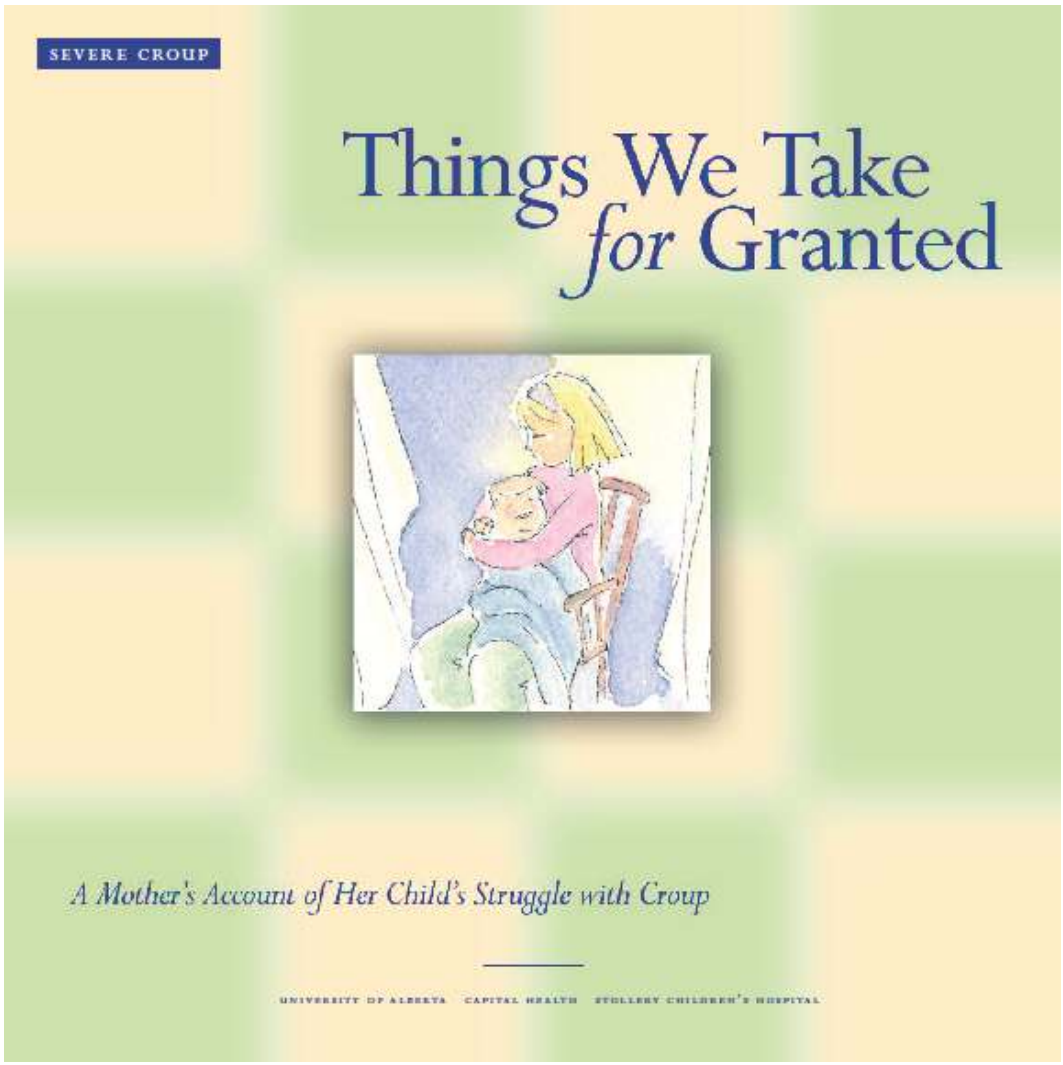
[Was It The Right Thing To Do?]

CROUP IS A VIRAL INFECTION of the windpipe (*trachea*) and vocal cords (*larynx*). It can affect children of all ages, but most commonly occurs in the first three years of life. The infection causes the lining of the throat and larynx to become red and swollen. In most cases, croup sounds worse than it actually is. Sometimes, the child may become very tired because of the extra work it takes to breathe. In very severe cases, the child's breathing can become blocked. Croup occurs most often in late fall, but can occur during any season, including summer.

Booklet sponsored by CIHR Team in Pediatric Emergency Medicine.

Capital Health

BOOK 2


SEVERE CROUP
Things We Take for Granted
A Mother's Account of Her Child's Struggle with Croup

A Message from Dr. William Craig

CROUP IS AN ILLNESS THAT AFFECTS YOUNG CHILDREN. It can come on quite suddenly and can cause severe coughing. In some cases, the child may have difficulty breathing. It is a stressful time for the parents of a young child.

Our first goal as health care professionals is to take care of your child. Another goal is to make sure that you have the information you need to understand your child's illness. Understanding the illness and how it is treated will ease the stress that you may have when your child is sick.

This booklet includes the story of a parent like you who had a child with croup. The story tells about one family's experience. The story also includes medical information on the illness and how it is treated.

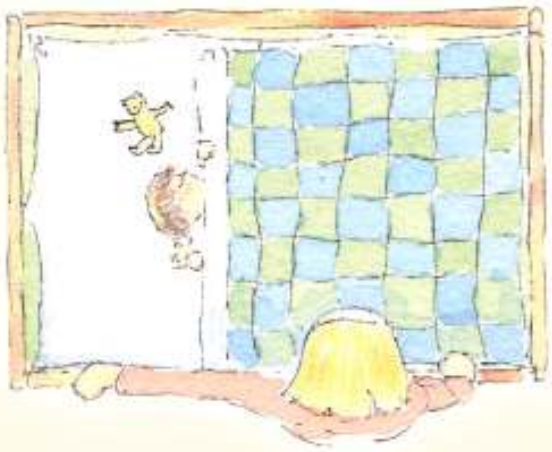
A story told through the eyes of a parent is a novel approach to passing on information. Most of us, from the very young to the very old, enjoy a good story. Stories also help us remember details – more so than reading a textbook or a scientific report.

This booklet includes one family's story but not all cases of croup are the same. Some are very mild and can be treated at home. In other cases the child should be seen by a doctor or at the emergency department.

I hope that this story will help answer some of the questions you have about croup. I also hope that you will find interest in reading other parents' stories, and that you will find comfort in knowing that you are not alone in your experience with croup. ■

DR. WILLIAM CRAIG, MD
Director of Pediatric Emergency,
Stollery Children's Hospital, Edmonton, AB

For more information on croup and how it can be treated please visit the Alberta Medical Association website to find the Clinical Practice Guideline (CPG) on Croup: ***www.albertadoctors.org***



I have just come from putting my 13-month old baby to bed for the night. I took extra time tonight to rock him to sleep. I listened to the sound of his breathing. I watched the steady movement of his chest as he breathed in and out. After what we have been through this last week, I find that I want to enjoy the small things we often take for granted.


PLEASE
TAKE
A
NUMBER

IT ALL STARTED A WEEK AGO when Matthew got a cough. The family doctor thought that it was just a regular cold.

"Give him Tylenol if he is uncomfortable, and make sure that he drinks lots of liquids," the doctor offered.

That night Matthew's coughing got worse. He also sounded out of breath and neither of us managed to get much sleep.

The next morning, I took him to a walk-in clinic. The doctor listened to Matthew's chest and told me that he had asthma. He gave us a prescription for some medicine called Ventolin. Even with the medicine, Matthew didn't seem to get any better. By suppertime his cough had a bark-like sound that started to worry me. I gave him another dose of the medicine and put him to bed right after dinner. A couple of hours later, Matthew was awake again. Now he was having a hard time breathing.

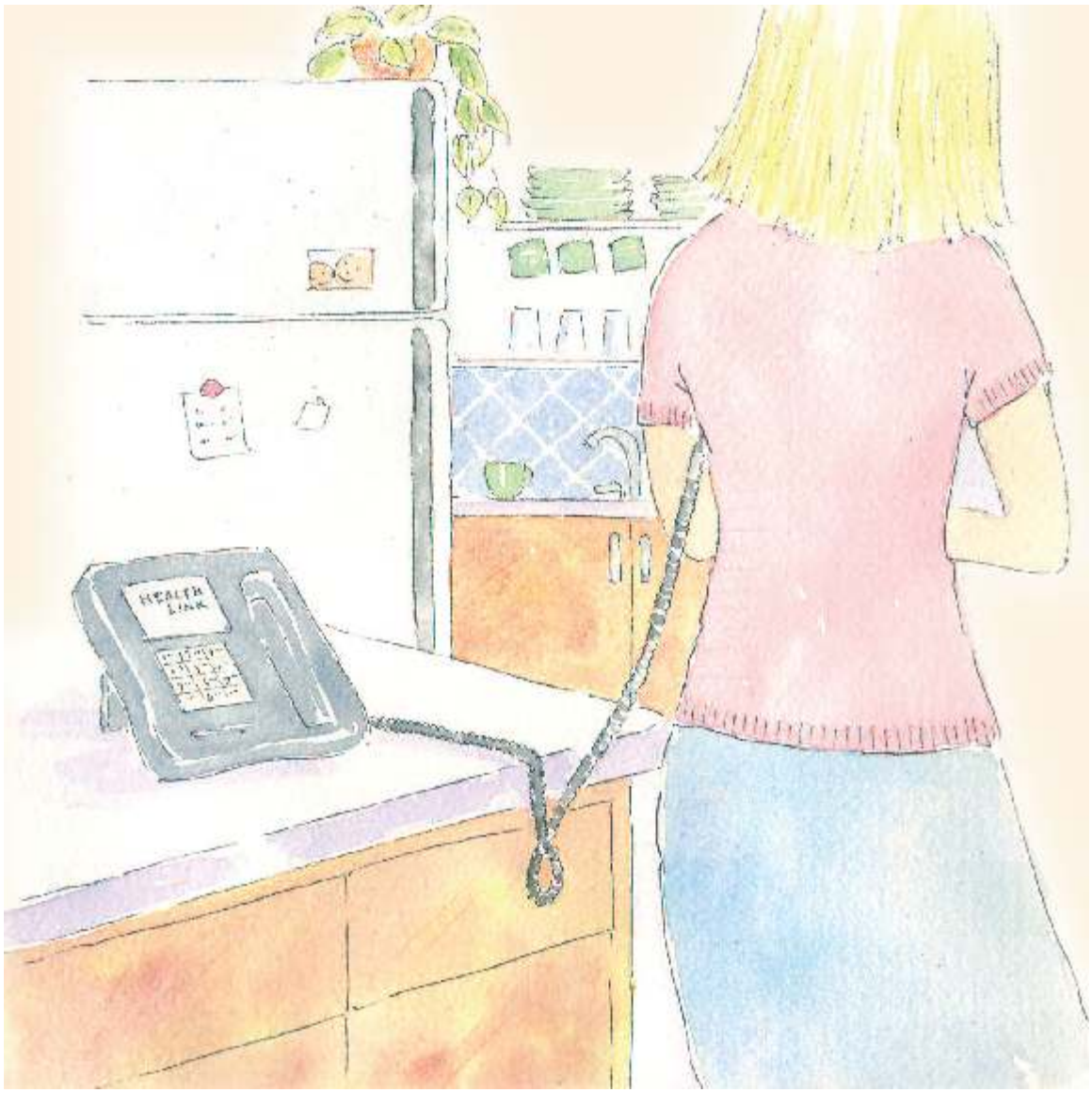
By suppertime his cough had a bark-like sound that started to worry me.

I remembered seeing a sign with a phone number to call for health information when I had last taken Matthew to the clinic for his shots. I found the number in the blue pages of the phone book under Health Link. Before long a nurse was on the other end of the line. The nurse asked a lot of questions about Matthew's cough and his breathing.

The most important thing to do for the coughing, is to have your child breathe cool air.

"The most important thing to do for the coughing," the nurse told me, "is to have your child breathe cool air. You can open a window in his room, take him outside for several minutes, take him for a car ride with the windows rolled down, or open the freezer door and let him breathe the cold air. But," she warned, "remember to keep him dressed warmly."

Then she added, "If you have tried these things and he is still having trouble breathing, you should take your son to the emergency department."


HEALTH
LINK

I opened the window, bundled him up in his favourite blanket, and we sat together in the rocking chair breathing in the cool air. The only thing that happened was that we both got cold, I got more and more nervous, and Matthew just wouldn't sit still. Next stop – I thought to myself as I packed his diaper bag – the emergency department.

Matthew, who was now feverish, struggled with every breath in and coughed with every breath out.

The nurse at the emergency department took us to an examination room right away. Matthew, who was now feverish, struggled with every breath in and coughed with every breath out. We had not waited long before the doctor came in. She read the chart and asked me some questions.

She listened to Matthew's chest and announced, "Your son has a bad case of croup."

I was confused. First I had been told that Matthew just had a cold. Then the doctor at the clinic said he had asthma. Now I was being told that he had croup. Which one did he have, if any? Why didn't anyone agree? Was he going to be okay?

"Croup is often mistaken for asthma, especially in young children", the doctor explained, "but croup affects the throat and the windpipe instead of the lungs. With croup we often hear a sharp, barky cough and sometimes we can hear a high pitched sound when the child breathes in. This sound is called stridor. When a child has asthma we hear a wheezing sound when they breathe out. This is one way that we can tell the difference between croup and asthma."

She explained that the medicine that I had been giving Matthew at home would not help with croup.

She explained that the medicine that I had been giving Matthew at home would not help with croup. Instead, she said that the nurse would give him a drug called epinephrine through a mask, and that this would help him breathe. She explained that the

The steroid would help with the swelling in his throat and make it easier for him to breathe.

epinephrine helps right away but doesn't last very long, so they would also give him a steroid called dexamethasone. The steroid would help with the swelling in his throat and make it easier for him to breathe, but it would take a few hours to work.

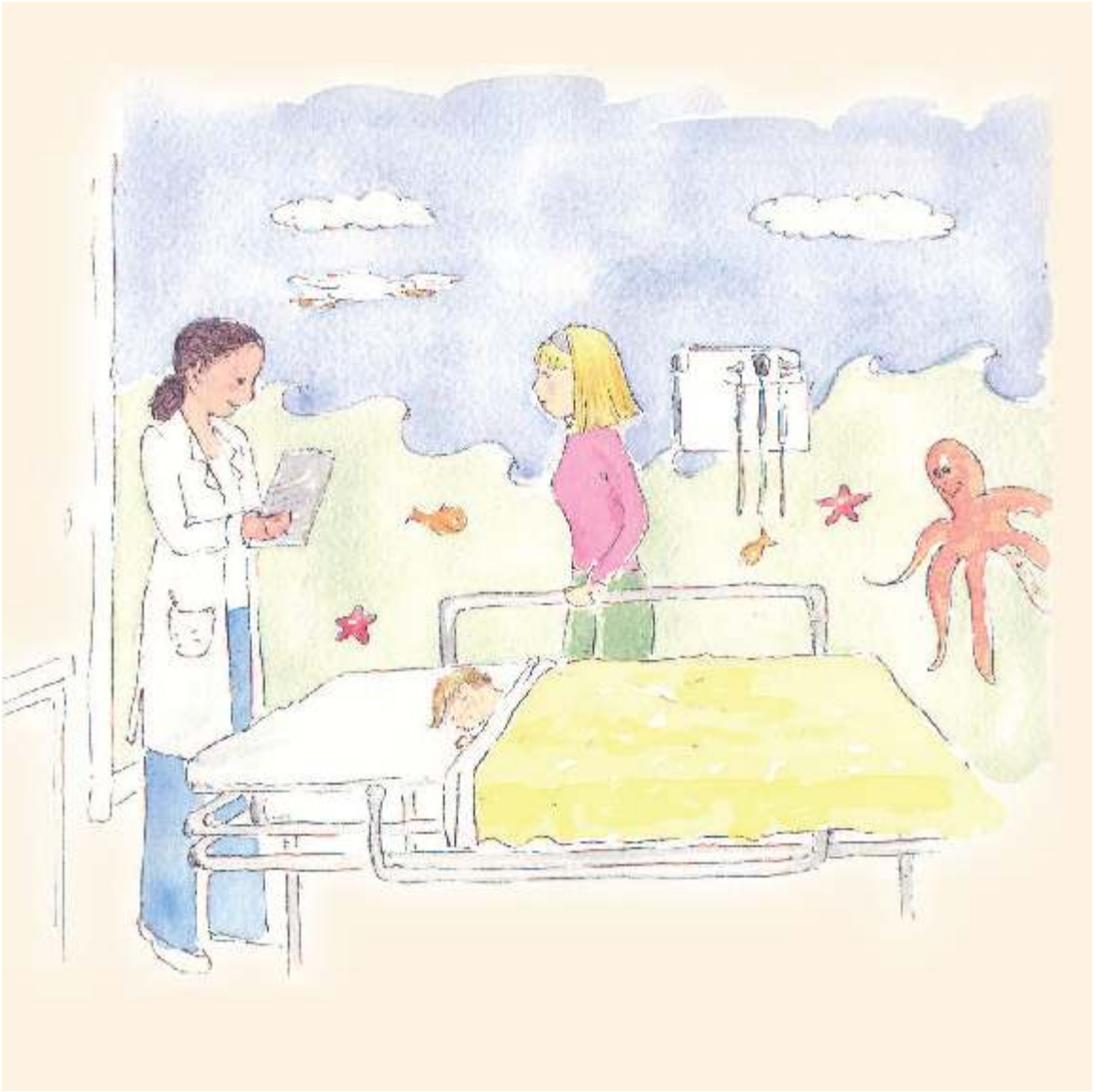
The nurse put the mask over Matthew's nose and mouth. Matthew became very upset. He was crying and trying to pull the mask off. I held the mask in place for several minutes and tried to comfort him. Matthew's breathing became much easier. Then the nurse lifted the mask and quickly squirted the dexamethasone syrup into his mouth with a syringe.

Shortly after the medicine in the mask, Matthew's breathing became almost normal. I hoped that we would be able to go home soon but the nurse explained that the effects of the medicine can wear off and that it was important to stay so that they could watch Matthew. Two hours later, Matthew was restless and coughing again. I was very upset when the doctor came in a while later and told us that we had to stay at the hospital.

I wondered whether any of the doctors knew what they were talking about.

"Sometimes the medicines don't work as well as we'd like." The nurse came in again and gave Matthew more epinephrine. The night drifted on...and the coughing continued.

At one point, the nurse poked her head into the room and asked if everything was OK. "NO", I wanted to shout, "IT'S NOT OK...My son is lying here in a hospital bed with a mask over his face like a patient on *ER*...I'm confused, I'm scared, and I'm tired." But I didn't speak the words that were circling in my head. I just nodded and forced out a smile.



Early the next morning, the nurse came in and said they had a bed ready for Matthew in the children's ward upstairs. Matthew spent the next two days in the hospital. They gave Matthew Tylenol a couple of times when the fever made him uncomfortable. Once when he was having a hard time breathing, he got the epinephrine through the face mask and his breathing improved right away.

Matthew would continue to get better over the next few days.

Matthew was allowed to go home the second afternoon. The doctor explained that Matthew would continue to get better over the next few days. She told me to come back to the hospital right away if Matthew got worse again.

It took three more days for Matthew's breathing to return to normal and for the coughing to stop.

I watched him closely from the rocking chair in his room...day after day, night after night... watching the steady rise and fall of his chest. The regular breathing is a comfort to me now...a sign that all is well.

Understanding Croup

Croup is an illness that affects a child's breathing. It is caused by many different viruses. It most often occurs in the fall and winter months.

Croup occurs most commonly in children between 6 months and 3 years of age, but can occur in children of all ages.

Croup is characterized by a barky cough that can start quite suddenly. Often the child will have a hoarse voice and difficulty breathing. You may hear a high pitched sound when your child breathes in – this is called stridor.

Croup is always worse at night or when your child is lying flat.

Croup usually gets worse on the second night of the illness, and lasts up to a week.

Antibiotics do not work on Croup because the infection is caused by a virus.

Your child may get croup by coming into contact with another person with the virus. It is spread through coughing, sneezing or contact with the mucous on tissues, toys or hands.

A doctor will assess how serious your child's croup case is. A mild case of croup means that the child has an occasional barky cough but no stridor is heard. In a moderate case, the child will have a frequent barky cough and stridor is easily heard when the child is calm. The child may or may not be agitated. When the illness is severe, the child will have a frequent barky cough, stridor can be easily heard and the child will be very distressed and agitated.

Things We Take *for* Granted

A Mother's Account of Her Child's Struggle with Croup

Capital Health

Booklet sponsored by CIHR Team in Pediatric Emergency Medicine.

BOOK 3


A Resourceful Father
and His Internet Connections
MILD CROUP
> Managing Croup at Home <
UNIVERSITY OF ALBERTA CAPITAL HEALTH STOLLERY CHILDREN'S HOSPITAL

> A Message from Dr. William Craig <

Croup is an illness that affects young children. It can come on quite suddenly and can cause severe coughing. In some cases, the child may have difficulty breathing. It is a stressful time for the parents of a young child.

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A story told through the eyes of a parent is a novel approach to passing on information. Most of us, from the very young to the very old, enjoy a good story. Stories also help us remember details – more so than reading a textbook or a scientific report.

This booklet includes one family's story but not all cases of croup are the same. Some are very mild and can be treated at home. In other cases the child should be seen by a doctor or at the emergency department. We have included information at the end of the booklet to help you in making the decision as to when you should see a doctor or go to the hospital.

I hope that this story will help answer some of the questions you have about croup. I also hope that you will find interest in reading other parents' stories, and that you will find comfort in knowing that you are not alone in your experience with croup.

Dr. William Craig, MD
Director of Pediatric Emergency
Stollery Children's Hospital
Edmonton, AB

Need Information?

For more information on Croup and how it can be treated please visit **www.albertadoctors.org**

> Managing Group at Home <

>>> Diane settled down in front of the TV to watch the ten o'clock news. She cherished this quiet time in her day after her two kids were in bed. Her husband Rick was finishing some work on the computer and would soon join her.

All of a sudden she heard cries coming from her daughter's room upstairs. Every once in a while, four-year old Emily would awaken after a bad dream, but this time it seemed different. Diane could sense real fear in her daughter's voice.

Diane heard Emily call out as she rushed up the stairs,

"Mommy, Mommy, I can't breathe!"


EMILY

> Emily began to cough as Diane opened the door. It was a bark-like cough that was incredibly loud.

Diane ran to the bed and tried to calm Emily down. They were sitting on the bed together when Rick appeared at the door.

"What's going on?" Rick asked.

"I'm not sure," Diane said. She thought about the day, trying to pinpoint a cause for Emily's sudden coughing and distress. Their day had been like any other. Emily had gone to playschool like she did regularly three times a week. She had eaten a good dinner and had gone to bed at her usual eight o'clock bedtime without trouble. Then she remembered. "I heard two mothers talking at the playschool. They said one of the little boys was sick at home with croup. Maybe Emily has the same thing."

"Diane, you stay with Emily, I want to see if I can find anything about croup on the Internet."

Diane rubbed her daughter's back and sang some of her favourite songs to calm her. Already Emily's breathing eased and she only coughed every once in a while.

Need Information?

Croup is an illness that affects a child's breathing. It is caused by many different viruses. It most often occurs in the fall and winter months but can happen in any season.

> Meanwhile Rick headed down the stairs and back to his computer. He opened up his favourite search engine and typed in "croup"...1,120,000 hits!

Need Information?

Croup occurs most commonly in children between 6 months and 3 years of age.

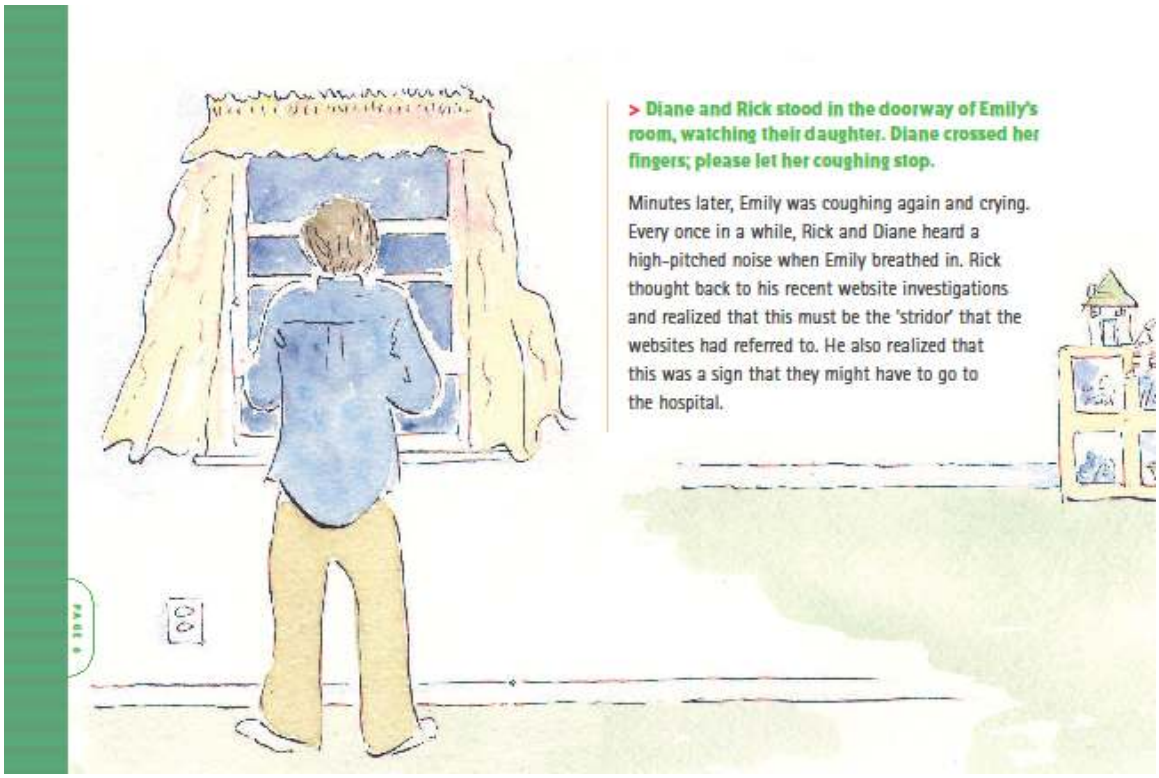
He spent twenty minutes looking at different websites, most of which gave him the same information. Then he headed back upstairs.

"Did you find anything?" Diane said as she settled Emily in bed.

"I expect you're right – Emily probably got croup from a little boy at playschool. One of the sites I read said that croup is spread through coughing and sneezing and germs can be picked up from tabletops, toys, or other shared objects."

"How can we be sure that it's croup and not something else?" Diane asked.

"I'm not sure, but the first website I checked said that croup is an infection that causes the windpipe and voice box to swell. The classic sign of croup is a loud, barky cough like the sound a seal makes. The child can show fast or difficult breathing and sometimes they make a squeaking sound when they breathe in."

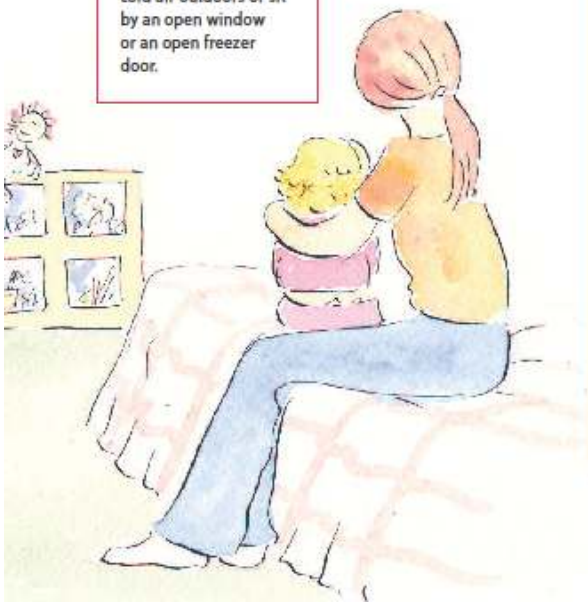


> **Diane and Rick stood in the doorway of Emily's room, watching their daughter. Diane crossed her fingers; please let her coughing stop.**

Minutes later, Emily was coughing again and crying. Every once in a while, Rick and Diane heard a high-pitched noise when Emily breathed in. Rick thought back to his recent website investigations and realized that this must be the 'stridor' that the websites had referred to. He also realized that this was a sign that they might have to go to the hospital.

Need Information?

Take your child into cold air outdoors or sit by an open window or an open freezer door.



> It was already late and Rick dreaded a long wait at the hospital. He thought back to the websites he had read and recalled the advice on some of them to have the child breathe in cool air.

"Let's try opening the window," Rick said. Diane was puzzled as to why they would open the window when their child was obviously sick. Rick told Diane about the information he had read on the Internet as he walked over and opened the window. They held a bedside vigil for another twenty minutes. Emily remained calm and the coughing became less frequent. They no longer heard the squeaking sound when she breathed in.

"Maybe that's done the trick," Rick said hopefully to Diane, "she already seems a lot better." They decided to close the window as they didn't want her to get chilled. "Why don't you head off to bed," Rick offered to Diane, "I'll stay with her while she falls asleep."

> Not long after Diane left the room, Emily's coughing started getting worse again and her breathing became strained. Rick realized that they might have to take Emily to the emergency department if things didn't get better soon.

He decided to try one more option that was suggested on several websites. He gathered up some pillows and a blanket and took Emily into the bathroom. He made a little nest for them on the floor with the pillows and blankets and began to run cool water from the shower. Over the next ten minutes, Emily's coughing eased. They sat for half an hour working through a pile of Emily's favourite bedtime storybooks. Somewhere between the fourth and fifth recounting of *Goodnight Moon*, Emily drifted off to sleep.

Rick bundled her up in the blanket and took her back to bed. He sat on the bed for some time to make sure that she was settled. When he was sure that she was sleeping soundly, Rick kissed Emily softly on the forehead and quietly left the room.

> **Though it was very late, Rick had a couple of last questions for the Internet. He quickly ran downstairs and picked up where he had left off. After a few minutes Diane walked in.**

"Yeah. Apparently kids can get croup more than once because there are many viruses that can cause croup. Kids can get croup when they become infected with each of these viruses."

"Did you find anything about how long Emily will have to stay at home?" Diane said as she pulled a chair up beside her husband.

Need Information?

Your child may get croup by coming into contact with another person with the virus. It is spread through coughing, sneezing or contact with the mucous on tissues, toys or hands.

"This site says that kids can go back to daycare or school when they no longer have a fever and they feel well enough to go back to their regular activities. It's okay to go back to daycare if she still has a cough."

Diane leaned forward in her chair, so she could get a better look at the computer screen. "How can we prevent Emily from getting croup again?"

"There's no vaccine or medicine to prevent croup, but we can decrease her chances of getting it again by making sure she washes her hands regularly, keeping her away from other people who are sick, and keeping her from sharing foods and drinks."

Need Information?

Hand washing, when done correctly, is the single most effective way to prevent the spread of communicable diseases. Good hand washing technique is easy to learn and can significantly reduce the spread of infectious diseases among both children and adults.

> **"It's been a long night," Rick said as he turned to Diane, "let's get to bed."**

Rick shut off the computer. As they made their way upstairs to bed they paused for a kiss and savored the quiet that had settled over the house. ■

Need Information?

Patients who have had prolonged high-pitch, noisy breathing should follow up with their family doctor or a doctor at the emergency department.

> Recommendations for Children with Croup <

- Keep your child and yourself calm. The symptoms can get worse if your child is excited.
- Take your child into cold air outdoors or sit by an open window or an open freezer door. (Remember to dress your child warmly.)
- A cool-mist humidifier may help.
- Sleep with window open.

- Give your child plenty of clear fluids or popsicles when awake.
- Encourage regular eating.
- Give your child acetaminophen (Tylenol or Tempra) or Ibuprofen (Advil or Motrin) for their fever or discomfort. Read the instructions on the bottle before you give your child any medication.

VISIT THE EMERGENCY DEPARTMENT IF:

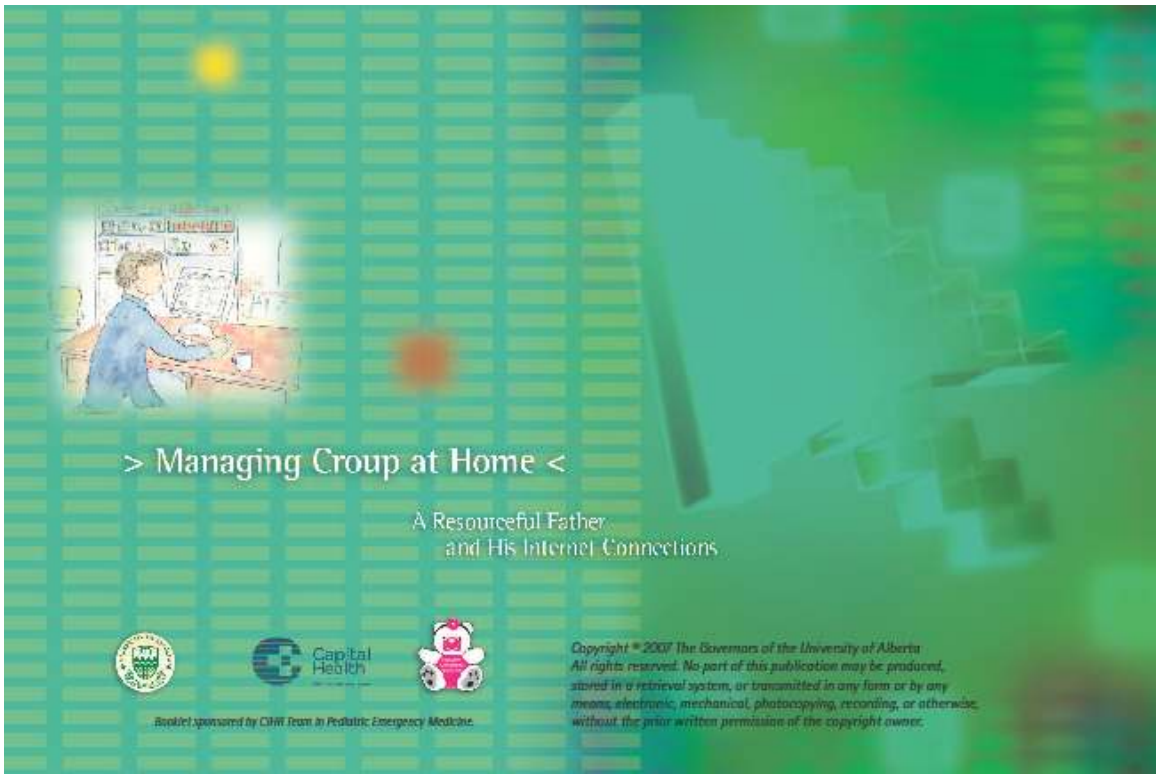
- Your child does not improve after 15 minutes of breathing cool air.
- Your child's breathing becomes difficult (when not coughing).
- Your child starts drooling or having a lot of trouble swallowing.
- Your child makes a high pitched noise when breathing in, even when they are calm, and the noise does not improve with cool air.
- Your child begins acting very sick.

CALL YOUR DOCTOR IF:

- Croup lasts more than 10 days
- Fever lasts more than 2 to 3 days
- You have other questions or concerns.

CALL 911 IMMEDIATELY IF YOUR CHILD:

- Has a bluish lip or fingernails.
- Appears very tired.
- Faints or passes out (unconscious).
- Stops breathing.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APPENDIX E. STANDARD INFORMATION SHEET

INSTRUCTIONS FOR PARENTS OF A CHILD WITH CROUP

What is Croup?

- Your child has croup which is caused by a virus that triggers swelling of the windpipe around the voice box. The swelling can cause a 'barky, seal-like' cough, a hoarse voice, and often a 'crowing' sound as your child breathes in. This sound is referred to as 'stridor'.
- The virus that causes croup is contagious. It is spread when your child coughs and breathes. In other family members – especially adults - this same virus can cause simple 'cold-like' symptoms such as hoarseness, cough, sore throat, and a runny nose.
- Your child's croupy cough will most likely disappear within a couple of days, though a few children continue to have a croupy cough for up to 7 days. Croup often disappears as quickly as it started, but in some cases, the harsh barky cough is followed by a loose cough and runny nose. Some children also develop ear infections.
- Croup is usually worse at night. Children who seemed well at bedtime can suddenly wake up with a barky cough and difficulty breathing. They often seem better during the day but then worsen again the next night.
- Croup recurs in some children but they usually "outgrow" the croup symptoms by ten years of age (though some not until they are teenagers.

What can I do to make my child more comfortable?

- If your child has a fever or a sore throat, you may give him or her acetaminophen (Tempra® or Tylenol®) or ibuprofen (Advil® or Motrin®). Doses are recommended on the side of the bottle, or ask a health care professional. **Never give your child more than 5 doses of acetaminophen or more than 4 doses of ibuprofen in a 24 hour period.**
- You can open your child's bedroom window a bit to let the cold air in, but remember to dress your child warmly. Don't worry - neither you nor your child will get sick from breathing cold air.
- Encourage 'cold' fluids such as juice, a slushy, or a Popsicle. Children with croup usually have a 'sore throat', and this may help to soothe it.
- If your child starts to make easily heard 'croupy sounds', and they are **NOT** 'blue in the face' or very restless with trouble breathing, try these 'home treatments':
  - In colder weather, bundle him/her up in warm clothes and take him or her outside in the colder air for 5 to 10 minutes.
  - In warmer weather, after making sure that your child is warmly dressed, open the freezer door and allow him or her to breath the cold air.
  - **Most importantly - if your child is upset - comfort him/her and speak calmly and in quiet tones. This will help more than anything to reduce breathing problems.**

How can I monitor my child to be sure they are okay?

- Croup is a 'noisy' disease, so you can check up on your child by always being within hearing range.
- Every once in a while watch and listen to your child breathing without a shirt or blanket covering their chest so that you can tell if they are having difficulty breathing, and need to be checked by a doctor:
  - Listen for a 'crowing sound' while your child is breathing in. If you hear this sound, note whether you hear it all the time, even when he/she is calm, or only when he/she is upset and crying.
  - Look to see whether your child's chest wall or the notch just below their 'Adam's Apple' is 'sucking' or 'caving in'.
  - See if you can get them to calm down or if they remain upset and restless even when you try to calm them
  - After making sure that you have enough light to see well, notice the coloring of your child's lips and face, checking for a 'bluish-grey' color.

The croup guideline for physicians and this patient hand-out were developed by a Clinical Practice Guideline working group which promotes appropriate, effective and quality medical care in Alberta. July 2003
This information is also available on the Alberta Medical Association web site:
www.albertadoctors.org

Should I call 911?

- **Call if:**
  - **Your child's face is bluish-grey in color for more than a few seconds; *or***
  - **Your child becomes unusually sleepy or 'glassy-eyed' while making croupy sounds; *or***
  - **Your child is really stressed, is struggling to breath, and you can not calm them within a few minutes.**
- **Remember that ambulance paramedics can start treatment for your child immediately, so that, if your child has very severe symptoms, it is safer to call '911' than to drive to the nearest hospital in your car.**

Should I seek medical care right away?

- Seek care right away if - after exposing your child to cold air:
  - Your child makes a persistent, easily heard 'crowing sound' with breathing.
  - Your child's chest wall 'sucks in' or 'caves in' as they breath.
  - Your child continues to have croupy symptoms that cause them to be significantly agitated or restless.
- When getting ready to go to the emergency department (or your doctor's), remember to dress both you and your child warmly, and - if it is not too cold outside - roll down your car window a bit. Breathing the cold air improves children's croupy symptoms, so that your child will most likely be quite a bit better when you arrive at the emergency department (or your doctor's office).

What medical treatment improves croup?

- Because a virus causes croup, antibiotics do not help.
- Anti-histamines and decongestants (over-the-counter 'cold' medications) DO NOT improve croup symptoms.
- 'Mist' therapy has been used for many years but it has never actually been shown to help improve croup symptoms.
- The most effective treatment for croup is dexamethasone, a kind of corticosteroid. Usually only one dose given by mouth is necessary. This medication, which is very safe, helps to reduce breathing troubles, reduces the chances that your child will need to come into hospital or return for medical care. This medicine starts to works within 2 or 3 hours, and lasts for a couple of days.
- Another effective treatment is an adrenaline (epinephrine) breathing mask, which works within minutes but lasts less than two hours. This is usually used only in children with more severe symptoms.

Is it safe for my child to come home (or should they stay in the hospital)?

- Most children with croup have mild symptoms so that it is safe for your child to be at home while they get better.
- About one in 25 children (4%) with croup needs to be kept in hospital for a few days until their breathing improves. If your child has to stay in the hospital, they will be watched, and if their breathing becomes really hard they will be given more adrenaline masks.
- Of those children who have to stay in hospital, one in every 100 (1%) have so much problem breathing that they need to have a special breathing tube put down their windpipe to help them breath for a few days. If this is necessary, your child would be transferred to an Intensive Care Unit (ICU). Even children with the most severe symptoms almost always get completely better within one or two weeks, without any left over problems.

Can I prevent my child from getting croup?

- There is no way to prevent your child from getting croup but hand washing helps to stop the spread of the viral infection that causes croup.

APPENDIX F. QUESTIONNAIRES

Croup Communication Tools (CCT Study): Baseline Questionnaire

Study ID: _____ _____ _____ – _____ _____ _____
Site number Patient number

Child's Initials: _____ _____ _____
F M L

PART A: SELF-EVALUATION QUESTIONNAIRE

DIRECTIONS

We would ask that you complete the following questions as they relate to your feelings about being in the emergency department right now and having a child who is sick.

A number of statements which people have used to describe themselves are given below. Read each statement and then circle the appropriate number to indicate how you feel *right* now, that is, *at this moment*. There are no right or wrong answers. Do not spend too much time on any one statement but give the answer which seems to describe your feelings best.

1----------2----------3----------4
Not at all Somewhat Moderately so Very much so

1.	I feel calm.....	1	2	3	4
2.	I feel secure.....	1	2	3	4
3.	I am tense.....	1	2	3	4
4.	I feel strained.....	1	2	3	4
5.	I feel at ease.....	1	2	3	4

Note: This tool consists of 20 questions but copyright restrictions prohibit including the entire instrument in documents including dissertations.

Croup Communication Tools (CCT Study) : Baseline Questionnaire

Study ID: _____ _____ _____ – _____ _____ _____ (Site number / Patient number)

Participant Initials: _____ _____ _____ (F M L)

PART B – PARENTAL CONCERNS (administered by research nurse/assistant)

For the following items please state the number that best shows your level of concern or anxiety on a scale of 0 to 3, where 0 = not at all concerned or anxious and 3 = extremely concerned or anxious.

Not at all ---------- Extremely
0 1 2 3

How concerned are you **right now** *(repeat this introduction as necessary for questions 21 to 31)*:

21.	about the uncomfortable aspect of your child's cough……..	0	1	2	3	NR
22.	about the unusual sound or nature of the cough……	0	1	2	3	NR
23.	about the unusual sound of your child's breathing……	0	1	2	3	NR
24.	about the effort that your child is making to breathe……	0	1	2	3	NR
25.	that your child is not getting enough oxygen……..	0	1	2	3	NR
26.	that your child may be wheezing or have asthma……..	0	1	2	3	NR
27.	that your child's sleep was disturbed……	0	1	2	3	NR
28.	that you felt increasingly tense or frustrated as a result of the illness……	0	1	2	3	NR
29.	that your child might be hospitalized……	0	1	2	3	NR
30.	that this illness might recur in the future……	0	1	2	3	NR
31.	about not knowing about this illness……..	0	1	2	3	NR
32.	On a scale of 1-10, with 1 being not very concerned and 10 being extremely concerned, how would you rate the overall concern you are feeling right now regarding your child's illness……	_____				NR

PART C – HOUSEHOLD AND HISTORY (administered by research nurse/assistant)

I have a few questions regarding your household and your child's medical history.

33) How many adults are presently living in the home? _____ —circle NR if no response

34) How many adults participate in the care of the child? _____ NR

35) What is the total number of children living in the home? _____ NR

36) When did you first notice respiratory symptoms in your child (i.e., runny nose, cough, noisy breathing)?
_____ / _____ / _____
mm dd yyyy

37) Has this child ever had croup before? Have your other children had croup?
_____ No history
_____ History this child
_____ History other child
_____ History both
_____ NR

38) Has this or another child ever been admitted to the hospital for croup before? This means staying overnight.
_____ No admits
_____ ED visit only this child
_____ ED visit only other child
_____ Previous admissions this child
_____ Previous admissions other child
_____ NR

39) Has this child, or any of your other children, ever been admitted to the intensive care unit for croup?
_____ No ICU admits
_____ ICU this child
_____ ICU other child
_____ NR

40) Has this child, or any of your other children, ever had a tube down his/her throat to help him breathe, for any reason?
_____ No history
_____ History this child
_____ History other child
_____ History both
_____ NR

41) Is there a history of previous serious illness for this child or do they have a chronic medical condition? (for example: asthma, pneumonia…)
_____ No
_____ Yes, details:
_____ NR

PART D – DEMOGRAPHIC INFORMATION (administered by research nurse/assistant)

[Interviewer to complete regarding participant (not child): male _____ female _____]

42) What year were you born? 19_____—circle NR if no response

43) What is your relationship to the child?

_____ Parent
_____ Step-parent
_____ Grandparent
_____ Other, specify: _____
_____ NR

44) What is the highest grade or year of school you completed?

_____ Grades 1-9
_____ Grades 10-11/Some high school
_____ High school graduate
_____ Some college/university
_____ College graduate
_____ Post-graduate education or degree
_____ NR

45) What is your marital status?

_____ Never married (single)
_____ Married/Common-law
_____ Separated, divorced or widowed
_____ Other, specify:
_____ NR

46) What is your household income per year?

_____ Less than $15,000 per year
_____ $15-29,000
_____ $30-44,000
_____ $45-59,000
_____ $60-74,000
_____ $75-90,000
_____ Over $90,000
_____ NR

47) Do you identify with an ethnic or minority group? _____no _____ yes (specify below) _____ NR

_____ First Nations
_____ Chinese
_____ South Asian (e.g., East Indian, Pakistani, Sri Lankan, etc)
_____ Black
_____ Filipino
_____ Latin American
_____ Southeast Asian (e.g., Vietnamese, Cambodian, Malaysian, Laotian, etc)
_____ Arab
_____ West Asian (e.g., Iranian, Afghan, etc)
_____ Korean
_____ Japanese
_____ Other, specify: _____

48) What was your place of birth? _____—circle NR if no response

48-a) If outside of Canada, how many years have you lived in Canada? ____—circle NR if no response

PART E – CROUP SEVERITY (assessed by research nurse/assistant)

Westley Croup Score*		
Item	**Category**	**Score (circle)**
Stridor	None	0
	When agitated	1
	At rest	2
Retractions	None	0
	Mild	1
	Moderate	2
	Severe	3
Air entry	Normal	0
	Decreased	1
	Markedly decreased	2
Cyanosis	None	0
(on room air)	With agitation	4
	At rest	5
Level of consciousness	Normal	0
	Disoriented	5
TOTAL SCORE	Time of assessment (hh:mm – 00:00 to 23:59): _____ : _____	

PART F: RESEARCH NURSE/ASSISTANT TO DOCUMENT FROM CHART

First three digits of postal code: ___ ___ ___

Child's date of birth (mm/dd/yyyy): _____ / _____ / _____

Date of ED visit (mm/dd/yyyy): _____ / _____ / _____

Time of triage (hh:mm – 00:00 to 23:59): _____ : _____

Patient disposition (site coordinator may have to complete this the following day):

- left without being seen ☐
- discharged home ☐
- admitted ☐
- other ☐ - specify:

Prior to recruitment patient was seen by (check all that apply):

- triage nurse ☐
- staff nurse ☐
- resident ☐
- staff physician ☐
- other ☐ – specify:

Prior to recruitment treatment was ordered by (check all that apply and specify who ordered which treatment below):

- triage nurse ☐
- staff nurse ☐
- resident ☐
- staff physician ☐
- other ☐ – specify:

Specify treatment given and time of treatment (hh:mm – 00:00 to 23:59) (check if not applicable ☐):

Croup Communication Tools (CCT Study): Discharge Questionnaire

Study ID: _____ – _____ Participant Initials: _____ _____ _____
Site number Patient number F M L

SELF-EVALUATION QUESTIONNAIRE

We would ask that you complete the following questions as they relate to your feelings about being in the emergency department right now and having a child who is sick.

A number of statements which people have used to describe themselves are given below. Read each statement and then circle the appropriate number to indicate how you feel *right* now, that is, *at this moment*. There are no right or wrong answers. Do not spend too much time on any one statement but give the answer which seems to describe your feelings best.

1----------2----------3----------4
Not at all Somewhat Moderately so Very much so

1.	I feel calm	1	2	3	4
2.	I feel secure	1	2	3	4
3.	I am tense	1	2	3	4
4.	I feel strained	1	2	3	4
5.	I feel at ease	1	2	3	4

Note: This tool consists of 20 questions but copyright restrictions prohibit including the entire instrument in documents including dissertations

21. Have you had a chance to read the study material that we gave to you about croup?
No _____ Yes _____

22. Since coming to the hospital today, have you read any information about croup other than the study material that we gave you?
No _____ Yes _____ —if yes, please describe: _____

Croup Communication Tools (CCT Study): Day 1 Questionnaire

Study ID: _____ _____ _____ – _____ _____ _____ (Site number / Patient number)

Child's Initials: _____ _____ _____ (F M L)

Dates and Times of Contact Attempts

Date	Time	Research nurse/assistant initial

Date and Time Reached: _____ / _____ / _____ (mm / dd / yyyy)

_____ _____ : _____ _____ (hh:mm (00:00-23:59))

Is your child: _____ at home or _____ in hospital?

PART A

I would ask that you answer the following questions as they relate to your feelings about having to deal with another occurrence of croup in the future.

I will read a number of statements which people have used to describe themselves. For each statement, please indicate how you think you would feel if one of your children had croup again in the future. There are no right or wrong answers. Do not spend too much time on any one statement but give the answer which seems to describe your feelings best.

The responses are "not at all", "somewhat", "moderately so", or "very much so".

1----------2----------3----------4
Not at all Somewhat Moderately so Very much so

1. I will feel calm.....	1	2	3	4	NR
2. I will feel secure.....	1	2	3	4	NR
3. I will be tense.....	1	2	3	4	NR
4. I will feel strained.....	1	2	3	4	NR
5. I will feel at ease.....	1	2	3	4	NR

Note: This tool consists of 20 questions but copyright restrictions prohibit including the entire instrument in documents including dissertations.

PART B – ONGOING SYMPTOMS

21) In the past 24 hours, when your child breathes in does he/she make a noise? (play audiotape of characteristic stridor associated with croup)

No (no noise) 0 _____
Yes (only when upset, active or agitated) 1 _____
Yes (at rest or when quiet) 2 _____

22) In the past 24 hours, has your child had a cough?

No (skip to question 24, score 0 on question 23) _____
Yes (Go to question 23) _____

23) Is the cough barky or not barky? (play audiotape of child with characteristic barky cough)

Not barky (0) _____
Barky (1) _____

Daily TOP Score (Circle total of question 21 plus question 23) = 0 1 2 3

24) In the past 24 hours, how many hours of his or her regular sleep do you think your child missed due to his or her croup?

Record number of hours stated by parent/caregiver: _____hours;

check if unknown or no response ☐

PART C – DECISION REGRET

The next set of questions is about your decision to take your child to the emergency department when they had croup. Please rate your agreement with the following statements. The responses are "strongly agree", "agree", "neither agree nor disagree", or "strongly disagree".

25) It was the right decision.
_____ strongly agree
_____ agree
_____ neither agree nor disagree
_____ strongly disagree
_____ NR

26) I regret the choice that was made.
_____ strongly agree
_____ agree
_____ neither agree nor disagree
_____ strongly disagree
_____ NR

27) I would go for the same choice if I had to do it over again.
_____ strongly agree
_____ agree
_____ neither agree nor disagree
_____ strongly disagree
_____ NR

28) The choice did my child a lot of harm.
_____ strongly agree
_____ agree
_____ neither agree nor disagree
_____ strongly disagree
_____ NR

29) The decision was a wise one.
_____ strongly agree
_____ agree
_____ neither agree nor disagree
_____ strongly disagree
_____ NR

PART D – SATISFACTION

30) In terms of meeting your expectations for treatment and care, rate your satisfaction with your overall visit to the emergency department
_____ very satisfied
_____ somewhat satisfied
_____ neither satisfied nor dissatisfied
_____ somewhat dissatisfied
_____ very dissatisfied
_____ NR

31) In terms of meeting your expectations for information, rate your satisfaction with the information handout that you were given when you were at the emergency department
_____ very satisfied
_____ somewhat satisfied
_____ neither satisfied nor dissatisfied
_____ somewhat dissatisfied
_____ very dissatisfied
_____ NR

PART E – RESOURCE UTILIZATION

BEFORE GOING TO THE EMERGENCY DEPARTMENT ON *specify day and/or date they were enrolled in the study)*:

32) Did you see or contact a health care professional because of your child's croup symptoms (barky cough, noisy and/or difficulty breathing)?

No _____
Yes _____
NR _____

If yes, check all that apply:

- Doctor ☐ (check if they just phoned ☐ or if they went to the office ☐)

  If they went to the office: how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

- Other health professional or healer ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, specify type of health professional/healer: _____

  If checked, did you pay out of pocked for this care: no ___, yes ___ - what was the cost: $_____

- Health Link ☐

The next few questions ask about WHEN YOU WENT TO THE EMERGENCY DEPARTMENT ON (*specify day and/or date they were enrolled in the study*):

33) Was your child transported by ambulance?

No _____
Yes _____
NR _____

34) How far did you have to travel from your home to get to the emergency department?

_____ circle: km / miles, circle if NR

35) Did you have to pay for parking?

No _____
Yes _____ - specify how much: $ _____
NR _____

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

The next few questions ask about the time SINCE COMING HOME FROM THE EMERGENCY DEPARTMENT ON (*specify date they were enrolled in the study*).

36) Have you had to use an ambulance service because of your child's illness with croup?
No _____
Yes _____
NR _____

37) Have you seen or contacted another health care professional because of your child's croup symptoms (barky cough, noisy and/or difficulty breathing)?
No _____
Yes _____
NR _____

If yes, check all that apply:

- Doctor ☐ (check if they just phoned ☐ or if they went to the office ☐)

  If they went to the office: how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

- Emergency department ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, was your child admitted to hospital because of the croup symptoms: no ___, yes ___

  If checked, was your child taken to the ED by ambulance: no _____, yes _____

- Other health professional or healer ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, specify type of health professional/healer: _____

  If checked, did you pay out of pocked for this care: no ___, yes ___ - what was the cost: $_____

- Health Link ☐

38) Since coming home from the emergency department on (*specify date that they were enrolled in the study*), has your child been admitted to hospital?
No _____
Yes _____ —If yes, has your child been discharged ☐ or are they still in hospital ☐?
If discharged, how many days were they in hospital?
NR _____

PART F – IMPACT ON FAMILY/CAREGIVER AND COSTS

DO NOT ASK QUESTIONS ON THIS AND THE NEXT PAGE IF CHILD IS STILL IN HOSPITAL (go to page 10)

The next set of questions ask about the impact of the illness on your family and costs associated with the illness. For each question, I would like you to think about the time BEFORE GOING to the emergency department and the time SINCE COMING HOME from the emergency department.

Before going to the ED	Since coming home from the ED
39) How many hours of sleep did you or your partner miss because of your child's illness with croup?	
_____ hours _____ NR	_____ hours _____ NR
40) How many hours of work did you or your partner miss because of your child's illness with croup?	
_____ hours _____ NR	_____ hours _____ NR
41) How many hours of housework did you or your partner miss because of your child's illness with croup?	
_____ hours _____ NR	_____ hours _____ NR
42) How many hours of your regular recreational activities did you or your partner miss because of your child's illness with croup?	
_____ hours _____ NR	_____ hours _____ NR
43) Do you have any other children who are school-aged?	
No _____ Yes _____ - did they miss any school: no _____, yes _____ - how much: NR _____	No _____ Yes _____ - did they miss any school: no _____, yes _____ - how much: NR _____
44) Did a physician prescribe any medicine because of your child's illness with croup before going to the emergency department?	
No _____ Yes _____ - provide details: What medication was prescribed? _____ How much did it cost? _____ NR _____	No _____ Yes _____ - provide details: What medication was prescribed? _____ How much did it cost? _____ NR _____

45) Did you have any expenses related to your child's illness with croup?	
No _____ Yes _____ - specify below NR _____	No _____ Yes _____ - specify below NR _____
-non prescription medications ☐ If checked, ask about the type of medications and costs:	-non prescription medications ☐ If checked, ask about the type of medications and costs:
-other supplies, such as a humidifier ☐ specify: If checked, ask about what it was, how many and the cost (e.g., 1 humidifier costing $35)	-other supplies, such as a humidifier ☐ specify: If checked, ask about what it was, how many and the cost (e.g., 1 humidifier costing $35)
-babysitting or childcare (more than usual) ☐ If checked, ask about number of hours and costs	-babysitting or childcare (more than usual) ☐ If checked, ask about number of hours and costs
-other, specify (ask as many details as possible about what it was, quantity and costs):	-other, specify (ask as many details as possible about what it was, quantity and costs):

ONLY ASK QUESTIONS ON THIS AND THE NEXT PAGE IF CHILD IS IN HOSPITAL.

The next set of questions ask about the impact of the illness on your family and costs associated with the illness. For each question, I would like you to think about the time BEFORE GOING to the emergency department and the time SINCE YOUR CHILD WAS ADMITTED TO HOSPITAL on (*specify day and/or date they were enrolled in the study*).

Before going to the ED	**Since your child was admitted to hospital**
46) How many hours of sleep have you or your partner missed because of your child's illness with croup?	
_____ hours _____ NR	_____ hours _____ NR
47) How many hours of work did you or your partner miss because of your child's illness with croup?	
_____ hours _____ NR	_____ hours _____ NR
48) How many hours of housework did you or your partner miss because of your child's illness with croup?	
_____ hours _____ NR	_____ hours _____ NR
49) How many hours of your regular recreational activities did you or your partner miss because of your child's illness with croup?	
_____ hours _____ NR	_____ hours _____ NR
50) Do you have any other children who are school-aged?	
No _____ Yes _____ - did they miss any school: no _____, yes _____ - how much: NR _____	No _____ Yes _____ - did they miss any school: no _____, yes _____ - how much: NR _____
51) Did a physician prescribe any medicine because of your child's illness with croup before going to the emergency department?	
No _____ Yes _____ - provide details: What medication was prescribed? _____ How much did it cost? _____ NR _____	

52) Have you had any expenses related to your child's illness with croup?

No _____
Yes _____ - specify below
NR _____

-non prescription medications ☐
If checked, ask about the type of medications and costs:

-other supplies, such as a humidifier ☐ specify:
If checked, ask about what it was, how many and the cost (e.g., 1 humidifier costing $35)

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

No _____
Yes _____ - specify below
NR _____

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

PART H: FINAL QUESTIONS

53) Since you were at the emergency department, have you had a chance to read the study material about croup that we gave to you?

No _____
Yes _____, specify: read some of it _____
read all of it once _____
read some of it more than once _____
read all of it more than once _____
NR _____

54) Since you were at the emergency department, have you read any information about croup other than what we gave you?

No _____
Yes _____ If yes, please describe (if they mention internet, ask about which websites):

NR _____

55) Are you confident that you will know what to do next time, that is, if one of your children has symptoms of croup again in the future?

_____ very confident
_____ somewhat confident
_____ not at all confident
_____ NR

END OF CALL

Thank parent for participating and answering their questions. Let them know that you will call again in 2 days. Ask them if they have a preference for when they would like you to call.

Note preference here:

Croup Communication Tools (CCT Study): Day 3 Questionnaire

Study ID: _____ _____ _____ – _____ _____ _____ (Site number / Patient number)

Child's Initials: _____ _____ _____ (F M L)

Record day and date of last call: _____

Dates and Times of Contact Attempts

Date	Time	RA initial

Date and Time Reached: _____ / _____ / _____ (mm / dd / yyyy)

_____ _____ : _____ _____ (hh:mm (00:00-23:59))

Is your child: _____ at home or _____ in hospital?

PART A – ONGOING SYMPTOMS

1) In the past 24 hours, when your child breathes in does he/she make a noise? (play audiotape of characteristic stridor associated with croup)

   No (no noise) 0 _____
   Yes (only when upset, active or agitated) 1 _____
   Yes (at rest or when quiet) 2 _____

2) In the past 24 hours, has your child had a cough?

   No (skip to question 4, score 0 on question 3) _____
   Yes (Go to question 3) _____

3) Is the cough barky or not barky? (play audiotape of child with characteristic barky cough)

   Not barky (0) _____
   Barky (1) _____

Daily TOP Score (Circle total of question 1 plus question 3) = 0 1 2 3

4) Since the last phone call, how many hours of his or her regular sleep do you think your child missed due to his or her croup?

   Record number of hours stated by parent/caregiver: _____hours

   check if unknown or no response ☐

PART B – IMPACT OF EVENT SCALE

ASK QUESTIONS IN PART B ONLY IF THE CHILD SCORED 0 ON THE TOP SCORE FROM PART A. IF CHILD SCORED GREATER THAN 0, GO TO PART C.

On _____ (day/date), you brought your child to the emergency department with an acute case of croup. I will read a list of comments made by people during stressful life events. Please indicate how frequently these comments were true for you DURING YOUR CHILD'S ILLNESS WITH CROUP. If they did not occur during that time, respond 'not at all'. If they did occur, please choose 'rarely', 'sometimes', 'often'.

5.	I thought about it when I didn't mean to……..	not at all	rarely	sometimes	often	NR
6.	I avoided letting myself get upset when I thought about it or was reminded of it……	not at all	rarely	sometimes	often	NR
7.	I tried to remove it from memory……	not at all	rarely	sometimes	often	NR
8.	I had trouble falling asleep or staying asleep, because of pictures or thoughts about it that came into my mind…..	not at all	rarely	sometimes	often	NR
9.	I had waves of strong feelings about it……..	not at all	rarely	sometimes	often	NR
10.	I had dreams about it……	not at all	rarely	sometimes	often	NR
11.	I stayed away from reminders of it……..	not at all	rarely	sometimes	often	NR
12.	I felt as if it hadn't happened or it wasn't real……	not at all	rarely	sometimes	often	NR
13.	I tried not to talk about it……..	not at all	rarely	sometimes	often	NR
14.	Pictures about it popped into my mind……	not at all	rarely	sometimes	often	NR
15.	Other things kept making me think about it……	not at all	rarely	sometimes	often	NR
16.	I was aware that I still had a lot of feelings about it, but I didn't deal with them……	not at all	rarely	sometimes	often	NR
17.	I tried not to think about it……	not at all	rarely	sometimes	often	NR
18.	Any reminder brought back feelings about it……..	not at all	rarely	sometimes	often	NR
19.	My feelings about it were kind of numb……..	not at all	rarely	sometimes	often	NR

PART C – KNOWLEDGE/RECALL

I am going to ask you some questions about croup. There will be some true and some false answers but if you are not sure of the answer, please let me know by choosing 'unsure'.

20) Croup is most often caused by a virus.
True _____ False _____ Unsure _____

21) Stridor is a sharp, barky cough that children get when they have croup.
True _____ False _____ Unsure _____

22) The barky cough is caused by swelling of the voice box, windpipe and vocal cords.
True _____ False _____ Unsure _____

23) You should always take your child to the emergency department if they have a sharp, barky cough.
True _____ False _____ Unsure _____

24) The most important thing you can do for your child's cough is to have them breathe warm air.
True _____ False _____ Unsure _____

25) A child should take antibiotics when they have croup.
True _____ False _____ Unsure _____

26) Dexamethasone is a steroid that helps reduce the swelling in the child's throat and windpipe.
True _____ False _____ Unsure _____

27) Croup most often occurs during the summer months.
True _____ False _____ Unsure _____

28) Taking your child out at night when it's cool can make the croup symptoms worse.
True _____ False _____ Unsure _____

29) Once the doctor in the emergency department sends you and your child home, there is only a small chance that you will need to return.
True _____ False _____ Unsure _____

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

PART D – RESOURCE UTILIZATION

The next few questions ask about the time SINCE I LAST CALLED YOU on (*name the day and date).*

30) Have you had to use an ambulance service because of your child's illness with croup?
No _____
Yes _____
NR _____

31) Have you seen or contacted another health care professional because of your child's croup symptoms (barky cough, noisy and/or difficulty breathing)?
No _____
Yes _____
NR _____

If yes, check all that apply:

- Doctor ☐ (check if they just phoned ☐ or if they went to the office ☐)

  If they went to the office: how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

- Emergency department ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, was your child admitted to hospital because of the croup symptoms: no ___, yes ___

  If checked, was your child taken to the ED by ambulance: no _____, yes _____

- Other health professional or healer ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, specify type of health professional/healer: _____

  If checked, did you pay out of pocked for this care: no ___, yes ___ - what was the cost: $_____

- Health Link ☐

32) Since our last call, has your child been admitted to hospital because of croup?
No _____
Yes _____—If yes, has your child been discharged ☐ or are they still in hospital ☐?
If discharged, how many days were they in hospital?
NR _____

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

PART E – IMPACT ON FAMILY/CAREGIVER AND COSTS

The next set of questions ask about the time SINCE WE LAST SPOKE ON THE PHONE (*name day and date*).

33) How many hours of sleep have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

34) How many hours of work have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

35) How many hours of housework have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

36) How many hours of your regular recreational activities have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

37) Do you have any other children who are school-aged?
No _____
Yes _____ - have they missed any school: no _____, yes _____ - how much:
NR _____

38) Has a physician prescribed any medicine because of your child's illness with croup?
No _____
Yes _____ - provide details:
What medication was prescribed? _____
How much did it cost? _____
NR _____

39) Have you had any expenses related to your child's illness with croup?
No _____
Yes _____ - specify below.
NR _____

-non prescription medications ☐
If checked, ask about the type of medications and costs:

-other supplies, such as a humidifier ☐ specify:
If checked, ask about what it was, how many and the cost (e.g., 1 humidifier costing $35)

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

ONLY ASK QUESTIONS ON THIS PAGE IF CHILD IS IN HOSPITAL.

The next set of questions ask about the time SINCE WE LAST SPOKE ON THE PHONE (*specify day and date*).

40) How many hours of sleep have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

41) How many hours of work have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

42) How many hours of housework have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

43) How many hours of your regular recreational activities have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

44) Do you have any other children who are school-aged?
No _____
Yes _____ - have they missed any school: no _____, yes _____ - how much:
NR _____

45) Have you had any expenses related to your child's illness with croup?
No _____
Yes _____ - specify below.
NR _____

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

PART F: FINAL QUESTIONS

46) Since our last call on *(day)*, have you read the study material about croup that we gave to you?

No _____
Yes _____, specify: read some of it _____
read all of it once _____
read some of it more than once _____
read all of it more than once _____
NR _____

47) Since our last call on *(day)*, have you read any information about croup other than what we gave you?

No _____
Yes _____ If yes, please describe (if they mention internet, ask about which websites):

NR _____

PART G – FOLLOWUP

Ask this question only if the child scored 0 on the TOP Score from Part A.

We would like to invite you to participate in an additional telephone interview to help us better understand the types of information that parents need and how this information can be formatted so that it is most useful to parents. This interview would take 30 to 60 minutes and would be scheduled at your convenience. If you agree, you may or may not be contacted. If you agree, I will give your name and telephone number to another researcher who may call you to arrange a time for the interview. Would you be willing to participate?

No _____ Yes _____

PART H - END OF CALL

If the child scored 0 on the TOP Score: Explain to the parent that you will only need to contact them once more in a year's time. Thank them for participating.

If the child scored greater than 0 on the TOP Score: Explain to the parent that you will contact them to ask a few questions in another two days. Ask them if they have a preference for when they would like you to call. Note preference.

Croup Communication Tools (CCT Study): Day 5 Questionnaire

Study ID: _____ _____ _____ – _____ _____ _____ (Site number / Patient number)

Child's Initials: _____ _____ _____ (F M L)

Record day and date of last call: _____

Dates and Times of Contact Attempts

Date	Time	RA initial

Date and Time Reached: _____ / _____ / _____ (mm / dd / yyyy)

_____ _____ : _____ _____ (hh:mm (00:00-23:59))

Is your child: _____ at home or _____ in hospital?

PART A – ONGOING SYMPTOMS

1) In the past 24 hours, when your child breathes in does he/she make a noise? (play audiotape of characteristic stridor associated with croup)

   No (no noise) 0 _____
   Yes (only when upset, active or agitated) 1 _____
   Yes (at rest or when quiet) 2 _____

2) In the past 24 hours, has your child had a cough?

   No (skip to question 4, score 0 on question 3) _____
   Yes (Go to question 3) _____

3) Is the cough barky or not barky? (play audiotape of child with characteristic barky cough)

   Not barky (0) _____
   Barky (1) _____

   Daily TOP Score (Circle total of question 1 plus question 3) = 0 1 2 3

4) Since the last phone call, how many hours of his or her regular sleep do you think your child missed due to his or her croup?

   Record number of hours stated by parent/caregiver: _____hours

   check if unknown or no response ☐

PART B – IMPACT OF EVENT SCALE

ASK QUESTIONS IN PART B ONLY IF THE CHILD SCORED 0 ON THE TOP SCORE FROM PART A. IF CHILD SCORED GREATER THAN 0, GO TO PART C.

On _____ (day/date), you brought your child to the emergency department with an acute case of croup. I will read a list of comments made by people during stressful life events. Please indicate how frequently these comments were true for you DURING YOUR CHILD'S ILLNESS WITH CROUP. If they did not occur during that time, respond 'not at all'. If they did occur, please choose 'rarely', 'sometimes', 'often'.

5.	I thought about it when I didn't mean to……..	not at all	rarely	sometimes	often	NR
6.	I avoided letting myself get upset when I thought about it or was reminded of it……	not at all	rarely	sometimes	often	NR
7.	I tried to remove it from memory……	not at all	rarely	sometimes	often	NR
8.	I had trouble falling asleep or staying asleep, because of pictures or thoughts about it that came into my mind…..	not at all	rarely	sometimes	often	NR
9.	I had waves of strong feelings about it……..	not at all	rarely	sometimes	often	NR
10.	I had dreams about it……	not at all	rarely	sometimes	often	NR
11.	I stayed away from reminders of it……..	not at all	rarely	sometimes	often	NR
12.	I felt as if it hadn't happened or it wasn't real……	not at all	rarely	sometimes	often	NR
13.	I tried not to talk about it……..	not at all	rarely	sometimes	often	NR
14.	Pictures about it popped into my mind……	not at all	rarely	sometimes	often	NR
15.	Other things kept making me think about it……	not at all	rarely	sometimes	often	NR
16.	I was aware that I still had a lot of feelings about it, but I didn't deal with them……	not at all	rarely	sometimes	often	NR
17.	I tried not to think about it……	not at all	rarely	sometimes	often	NR
18.	Any reminder brought back feelings about it……..	not at all	rarely	sometimes	often	NR
19.	My feelings about it were kind of numb……..	not at all	rarely	sometimes	often	NR

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

PART C – RESOURCE UTILIZATION

The next few questions ask about the time SINCE I LAST CALLED YOU on (*name the day and date).*

20) Have you had to use an ambulance service because of your child's illness with croup?

No _____
Yes _____
NR _____

21) Have you seen or contacted another health care professional because of your child's croup symptoms (barky cough, noisy and/or difficulty breathing)?

No _____
Yes _____
NR _____

If yes, check all that apply:

- Doctor ☐ (check if they just phoned ☐ or if they went to the office ☐)

  If they went to the office: how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

- Emergency department ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, was your child admitted to hospital because of the croup symptoms: no ___, yes ___

  If checked, was your child taken to the ED by ambulance: no _____, yes _____

- Other health professional or healer ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, specify type of health professional/healer: _____

  If checked, did you pay out of pocked for this care: no ___, yes ___ - what was the cost: $_____

- Health Link ☐

22) Since our last call, has your child been admitted to hospital because of croup?

No _____
Yes _____—If yes, has your child been discharged ☐ or are they still in hospital ☐?
If discharged, how many days were they in hospital?
NR _____

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

PART D – IMPACT ON FAMILY/CAREGIVER AND COSTS

The next set of questions ask about the time SINCE WE LAST SPOKE ON THE PHONE (*name day and date*).

23) How many hours of sleep have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

24) How many hours of work have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

25) How many hours of housework have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

26) How many hours of your regular recreational activities have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

27) Do you have any other children who are school-aged?
No _____
Yes _____ - have they missed any school: no _____, yes _____ - how much:
NR _____

28) Has a physician prescribed any medicine because of your child's illness with croup?
No _____
Yes _____ - provide details:
What medication was prescribed? _____
How much did it cost? _____
NR _____

29) Have you had any expenses related to your child's illness with croup?
No _____
Yes _____ - specify below.
NR _____

-non prescription medications ☐
If checked, ask about the type of medications and costs:

-other supplies, such as a humidifier ☐ specify:
If checked, ask about what it was, how many and the cost (e.g., 1 humidifier costing $35)

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

ONLY ASK QUESTIONS ON THIS PAGE IF CHILD IS IN HOSPITAL.

The next set of questions ask about the time SINCE WE LAST SPOKE ON THE PHONE (*specify day and date*).

30) How many hours of sleep have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

31) How many hours of work have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

32) How many hours of housework have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

33) How many hours of your regular recreational activities have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

34) Do you have any other children who are school-aged?
No _____
Yes _____ - have they missed any school: no _____, yes _____ - how much:
NR _____

35) Have you had any expenses related to your child's illness with croup?
No _____
Yes _____ - specify below.
NR _____

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

PART E: FINAL QUESTIONS

36) Since our last call on *(day)*, have you read the study material about croup that we gave to you?

No _____
Yes _____, specify: read some of it _____
read all of it once _____
read some of it more than once _____
read all of it more than once _____
NR _____

37) Since our last call on *(day)*, have you read any information about croup other than what we gave you?

No _____
Yes _____ If yes, please describe (if they mention internet, ask about which websites):

NR _____

PART F – FOLLOWUP

Ask this question only if the child scored 0 on the TOP Score from Part A.

We would like to invite you to participate in an additional telephone interview to help us better understand the types of information that parents need and how this information can be formatted so that it is most useful to parents. This interview would take 30 to 60 minutes and would be scheduled at your convenience. If you agree, you may or may not be contacted. If you agree, I will give your name and telephone number to another researcher who may call you to arrange a time for the interview. Would you be willing to participate?

No _____ Yes _____

PART G - END OF CALL

If the child scored 0 on the TOP Score: Explain to the parent that you will only need to contact them once more in a year's time. Thank them for participating.

If the child scored greater than 0 on the TOP Score: Explain to the parent that you will contact them to ask a few questions in another two days. Ask them if they have a preference for when they would like you to call.

Croup Communication Tools (CCT Study): Day 7 Questionnaire

Study ID: _____ _____ _____ – _____ _____ _____ (Site number / Patient number)

Child's Initials: _____ _____ _____ (F M L)

Record day and date of last call: _____

Dates and Times of Contact Attempts

Date	Time	RA initial

Date and Time Reached: _____ / _____ / _____ (mm / dd / yyyy)

_____ _____ : _____ _____ (hh:mm (00:00-23:59))

Is your child: _____ at home or _____ in hospital?

PART A – ONGOING SYMPTOMS

1) In the past 24 hours, when your child breathes in does he/she make a noise? (play audiotape of characteristic stridor associated with croup)

   No (no noise) 0 _____
   Yes (only when upset, active or agitated) 1 _____
   Yes (at rest or when quiet) 2 _____

2) In the past 24 hours, has your child had a cough?

   No (skip to question 4, score 0 on question 3) _____
   Yes (Go to question 3) _____

3) Is the cough barky or not barky? (play audiotape of child with characteristic barky cough)

   Not barky (0) _____
   Barky (1) _____

Daily TOP Score (Circle total of question 1 plus question 3) = 0 1 2 3

4) Since our last call, how many hours of his or her regular sleep do you think your child missed due to his or her croup?

   Record number of hours stated by parent/caregiver: _____hours

   check if unknown or no response ☐

PART B – IMPACT OF EVENT SCALE

ASK QUESTIONS IN PART B ONLY IF THE CHILD SCORED 0 ON THE TOP SCORE FROM PART A. IF CHILD SCORED GREATER THAN 0, GO TO PART C.

On _____ (day/date), you brought your child to the emergency department with an acute case of croup. I will read a list of comments made by people during stressful life events. Please indicate how frequently these comments were true for you DURING YOUR CHILD'S ILLNESS WITH CROUP. If they did not occur during that time, respond 'not at all'. If they did occur, please choose 'rarely', 'sometimes', 'often'.

5.	I thought about it when I didn't mean to……..	not at all	rarely	sometimes	often	NR
6.	I avoided letting myself get upset when I thought about it or was reminded of it……	not at all	rarely	sometimes	often	NR
7.	I tried to remove it from memory……	not at all	rarely	sometimes	often	NR
8.	I had trouble falling asleep or staying asleep, because of pictures or thoughts about it that came into my mind…..	not at all	rarely	sometimes	often	NR
9.	I had waves of strong feelings about it……..	not at all	rarely	sometimes	often	NR
10.	I had dreams about it……	not at all	rarely	sometimes	often	NR
11.	I stayed away from reminders of it……..	not at all	rarely	sometimes	often	NR
12.	I felt as if it hadn't happened or it wasn't real……	not at all	rarely	sometimes	often	NR
13.	I tried not to talk about it……..	not at all	rarely	sometimes	often	NR
14.	Pictures about it popped into my mind……	not at all	rarely	sometimes	often	NR
15.	Other things kept making me think about it……	not at all	rarely	sometimes	often	NR
16.	I was aware that I still had a lot of feelings about it, but I didn't deal with them……	not at all	rarely	sometimes	often	NR
17.	I tried not to think about it……	not at all	rarely	sometimes	often	NR
18.	Any reminder brought back feelings about it……..	not at all	rarely	sometimes	often	NR
19.	My feelings about it were kind of numb……..	not at all	rarely	sometimes	often	NR

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

PART C – RESOURCE UTILIZATION

The next few questions ask about the time SINCE I LAST CALLED YOU on (*name the day and date).*

20) Have you had to use an ambulance service because of your child's illness with croup?

No _____
Yes _____
NR _____

21) Have you seen or contacted another health care professional because of your child's croup symptoms (barky cough, noisy and/or difficulty breathing)?

No _____
Yes _____
NR _____

If yes, check all that apply:

- Doctor ☐ (check if they just phoned ☐ or if they went to the office ☐)

  If they went to the office: how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

- Emergency department ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, was your child admitted to hospital because of the croup symptoms: no ___, yes ___

  If checked, was your child taken to the ED by ambulance: no _____, yes _____

- Other health professional or healer ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, specify type of health professional/healer: _____

  If checked, did you pay out of pocked for this care: no ___, yes ___ - what was the cost: $_____

- Health Link ☐

22) Since our last call, has your child been admitted to hospital because of croup?

No _____
Yes _____—If yes, has your child been discharged ☐ or are they still in hospital ☐?
If discharged, how many days were they in hospital?
NR _____

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

PART D – IMPACT ON FAMILY/CAREGIVER AND COSTS

The next set of questions ask about the time SINCE WE LAST SPOKE ON THE PHONE (*name day and date*).

23) How many hours of sleep have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

24) How many hours of work have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

25) How many hours of housework have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

26) How many hours of your regular recreational activities have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

27) Do you have any other children who are school-aged?
No _____
Yes _____ - have they missed any school: no _____, yes _____ - how much:
NR _____

28) Has a physician prescribed any medicine because of your child's illness with croup?
No _____
Yes _____ - provide details:
What medication was prescribed? _____
How much did it cost? _____
NR _____

29) Have you had any expenses related to your child's illness with croup?
No _____
Yes _____ - specify below.
NR _____

-non prescription medications ☐
If checked, ask about the type of medications and costs:

-other supplies, such as a humidifier ☐ specify:
If checked, ask about what it was, how many and the cost (e.g., 1 humidifier costing $35)

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

ONLY ASK QUESTIONS ON THIS PAGE IF CHILD IS IN HOSPITAL.

The next set of questions ask about the time SINCE WE LAST SPOKE ON THE PHONE (*specify day and date*).

30) How many hours of sleep have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

31) How many hours of work have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

32) How many hours of housework have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

33) How many hours of your regular recreational activities have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

34) Do you have any other children who are school-aged?
No _____
Yes _____ - have they missed any school: no _____, yes _____ - how much:

35) Have you had any expenses related to your child's illness with croup?
No _____
Yes _____ - specify below.
NR _____

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

PART E: FINAL QUESTIONS

36) Since our last call on *(day)*, have you read the study material about croup that we gave to you?

No _____
Yes _____, specify: read some of it _____
read all of it once _____
read some of it more than once _____
read all of it more than once _____
NR _____

37) Since our last call on *(day)*, have you read any information about croup other than what we gave you?

No _____
Yes _____ If yes, please describe (if they mention internet, ask about which websites):

NR _____

PART F – FOLLOWUP

Ask this question only if the child scored 0 on the TOP Score from Part A.

We would like to invite you to participate in an additional telephone interview to help us better understand the types of information that parents need and how this information can be formatted so that it is most useful to parents. This interview would take 30 to 60 minutes and would be scheduled at your convenience. If you agree, you may or may not be contacted. If you agree, I will give your name and telephone number to another researcher who may call you to arrange a time for the interview. Would you be willing to participate?

No _____ Yes _____

PART G - END OF CALL

If the child scored 0 on the TOP Score: Explain to the parent that you will only need to contact them once more in a year's time. Thank them for participating.

If the child scored greater than 0 on the TOP Score: Explain to the parent that you will contact them to ask a few questions in another two days. Ask them if they have a preference for when they would like you to call.

Croup Communication Tools (CCT Study): Day 9 Questionnaire

Study ID: _____ _____ _____ – _____ _____ _____ (Site number / Patient number)

Child's Initials: _____ _____ _____ (F M L)

Record day and date of last call: _____

Dates and Times of Contact Attempts

Date	Time	RA initial

Date and Time Reached: _____ / _____ / _____ (mm / dd / yyyy)

_____ _____ : _____ _____ (hh:mm (00:00-23:59))

Is your child: _____ at home or _____ in hospital?

PART A – ONGOING SYMPTOMS

1) In the past 24 hours, when your child breathes in does he/she make a noise? (play audiotape of characteristic stridor associated with croup)

   No (no noise) 0 _____
   Yes (only when upset, active or agitated) 1 _____
   Yes (at rest or when quiet) 2 _____

2) In the past 24 hours, has your child had a cough?

   No (skip to question 4, score 0 on question 3) _____
   Yes (Go to question 3) _____

3) Is the cough barky or not barky? (play audiotape of child with characteristic barky cough)

   Not barky (0) _____
   Barky (1) _____

   Daily TOP Score (Circle total of question 1 plus question 3) = 0 1 2 3

4) Since our last call, how many hours of his or her regular sleep do you think your child missed due to his or her croup?

   Record number of hours stated by parent/caregiver: _____hours

   check if unknown or no response ☐

PART B – IMPACT OF EVENT SCALE

ASK QUESTIONS IN PART B ONLY IF THE CHILD SCORED 0 ON THE TOP SCORE FROM PART A. IF CHILD SCORED GREATER THAN 0, GO TO PART C.

On _____ (day/date), you brought your child to the emergency department with an acute case of croup. I will read a list of comments made by people during stressful life events. Please indicate how frequently these comments were true for you DURING YOUR CHILD'S ILLNESS WITH CROUP. If they did not occur during that time, respond 'not at all'. If they did occur, please choose 'rarely', 'sometimes', 'often'.

5.	I thought about it when I didn't mean to……..	not at all	rarely	sometimes	often	NR
6.	I avoided letting myself get upset when I thought about it or was reminded of it……	not at all	rarely	sometimes	often	NR
7.	I tried to remove it from memory……	not at all	rarely	sometimes	often	NR
8.	I had trouble falling asleep or staying asleep, because of pictures or thoughts about it that came into my mind…..	not at all	rarely	sometimes	often	NR
9.	I had waves of strong feelings about it……..	not at all	rarely	sometimes	often	NR
10.	I had dreams about it……..	not at all	rarely	sometimes	often	NR
11.	I stayed away from reminders of it……..	not at all	rarely	sometimes	often	NR
12.	I felt as if it hadn't happened or it wasn't real……	not at all	rarely	sometimes	often	NR
13.	I tried not to talk about it……..	not at all	rarely	sometimes	often	NR
14.	Pictures about it popped into my mind……	not at all	rarely	sometimes	often	NR
15.	Other things kept making me think about it……	not at all	rarely	sometimes	often	NR
16.	I was aware that I still had a lot of feelings about it, but I didn't deal with them……	not at all	rarely	sometimes	often	NR
17.	I tried not to think about it……..	not at all	rarely	sometimes	often	NR
18.	Any reminder brought back feelings about it……..	not at all	rarely	sometimes	often	NR
19.	My feelings about it were kind of numb……..	not at all	rarely	sometimes	often	NR

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

PART C – RESOURCE UTILIZATION

The next few questions ask about the time SINCE I LAST CALLED YOU on (*name the day and date).*

20) Have you had to use an ambulance service because of your child's illness with croup?

No _____
Yes _____
NR _____

21) Have you seen or contacted another health care professional because of your child's croup symptoms (barky cough, noisy and/or difficulty breathing)?

No _____
Yes _____
NR _____

If yes, check all that apply:

- Doctor ☐ (check if they just phoned ☐ or if they went to the office ☐)

  If they went to the office: how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

- Emergency department ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, was your child admitted to hospital because of the croup symptoms: no ___, yes ___

  If checked, was your child taken to the ED by ambulance: no _____, yes _____

- Other health professional or healer ☐

  If checked, how far did you have to travel: _____ circle: km / miles

  Did you have to pay for parking: no _____, yes _____ - specify how much: $ _____

  If checked, specify type of health professional/healer: _____

  If checked, did you pay out of pocked for this care: no ___, yes ___ - what was the cost: $_____

- Health Link ☐

22) Since our last call, has your child been admitted to hospital because of croup?

No _____
Yes _____—If yes, has your child been discharged ☐ or are they still in hospital ☐?
If discharged, how many days were they in hospital?
NR _____

DO NOT ASK QUESTIONS ON THIS PAGE IF CHILD IS STILL IN HOSPITAL.

PART D – IMPACT ON FAMILY/CAREGIVER AND COSTS

The next set of questions ask about the time SINCE WE LAST SPOKE ON THE PHONE (*name day and date*).

23) How many hours of sleep have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

24) How many hours of work have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

25) How many hours of housework have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

26) How many hours of your regular recreational activities have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

27) Do you have any other children who are school-aged?
No _____
Yes _____ - have they missed any school: no _____, yes _____ - how much:
NR _____

28) Has a physician prescribed any medicine because of your child's illness with croup?
No _____
Yes _____ - provide details:
What medication was prescribed? _____
How much did it cost? _____
NR _____

29) Have you had any expenses related to your child's illness with croup?
No _____
Yes _____ - specify below.
NR _____

-non prescription medications ☐
If checked, ask about the type of medications and costs:

-other supplies, such as a humidifier ☐ specify:
If checked, ask about what it was, how many and the cost (e.g., 1 humidifier costing $35)

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

ONLY ASK QUESTIONS ON THIS PAGE IF CHILD IS IN HOSPITAL.

The next set of questions ask about the time SINCE WE LAST SPOKE ON THE PHONE (*specify day and date*).

30) How many hours of sleep have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

31) How many hours of work have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

32) How many hours of housework have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

33) How many hours of your regular recreational activities have you or your partner missed because of your child's illness with croup?
_____ hours
_____ NR

34) Do you have any other children who are school-aged?
No _____
Yes _____ - have they missed any school: no _____, yes _____ - how much:

35) Have you had any expenses related to your child's illness with croup?
No _____
Yes _____ - specify below.
NR _____

-babysitting or childcare (more than usual) ☐
If checked, ask about number of hours and costs

-other, specify (ask as many details as possible about what it was, quantity and costs):

PART E: FINAL QUESTIONS

36) Since our last call on *(day)*, have you read the study material about croup that we gave to you?

No _____
Yes _____, specify: read some of it _____
read all of it once _____
read some of it more than once _____
read all of it more than once _____
NR _____

37) Since our last call on *(day)*, have you read any information about croup other than what we gave you?

No _____
Yes _____ If yes, please describe (if they mention internet, ask about which websites):

NR _____

PART F – FOLLOWUP

We would like to invite you to participate in an additional telephone interview to help us better understand the types of information that parents need and how this information can be formatted so that it is most useful to parents. This interview would take 30 to 60 minutes and would be scheduled at your convenience. If you agree, you may or may not be contacted. If you agree, I will give your name and telephone number to another researcher who may call you to arrange a time for the interview. Would you be willing to participate?

No _____ Yes _____

PART G - END OF CALL

Explain to the parent that you will only need to contact them once more in a year's time. Thank them for participating.

APPENDIX G. SITE-SPECIFIC RESULTS

G-1. Alberta Children's Hospital

G-1.1 Study sample

Overall 137 parents were recruited at the Alberta Children's Hospital in Calgary: 70 participants were randomized to receive story booklets and 67 received standard information sheets. Figure G-1 describes the recruitment and follow-up of study participants to day 3 which was the last follow-up point required for all participants.

Characteristics of the trial participants are detailed in Tables G-1 to G-3. There were no notable differences between groups in terms of demographic variables (Table G-1).

Table G-2 presents the results for parental concern at baseline. Overall, parents demonstrated a moderate level of concern with a mean self-rating of 6.32 (SD 2.49) on a scale of 1 to 10, where 10 represents the highest level of concern. The items that generated the most concern were the unusual sound of the child's breathing (48.2% expressed extreme concern), the effort the child made to breathe (45.3% extreme concern), and the unusual sound or nature of the sough (40.9% extreme concern). There were no notable differences in overall or item-specific concern between study groups.

The majority of participants had no prior history of croup admissions, ICU admissions, or intubations. A substantial proportion (39%) of participants reported a previous experience with croup either with the same or another child, while 24% reported a prior serious illness or medical condition for their child. The most commonly reported serious illnesses/medical conditions were asthma (n=17), pneumonia (n=5), and complications of prematurity (n=5). Overall there were no differences between groups in the prevalence of previous medical history or experiences with participants' children.

The majority of patients presented with mild croup with a median croup score of 1 (IQR 1,3) on a scale of 0 to 17. 88% of the patients were discharged home from the ED while 9% were admitted (6 patients in each group). Only 8% had been seen by the staff physician before being recruited into the study. All of the patients were seen by a triage nurse prior to recruitment and 88% had been ordered treatment.

Approximately 56% of participants read the study material during their ED stay while 20% read additional information on croup. The groups differed somewhat with fewer parents in the story group reading the study material (46% versus 67%) but more parents in the story group reading additional material (24% versus 15%).

Figure G-1. Recruitment and follow-up of study participants

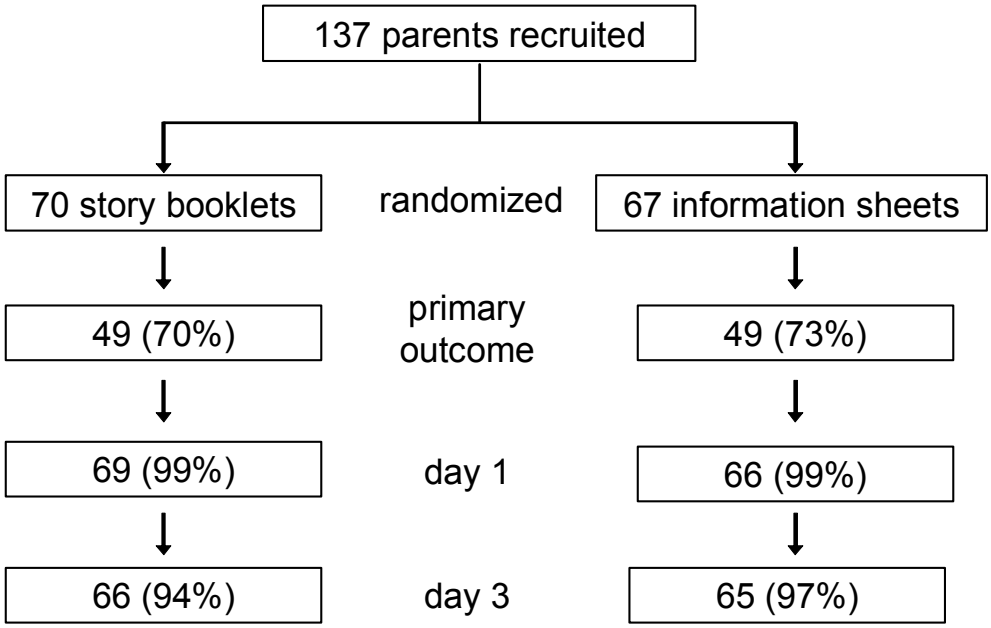


Table G-1. Demographics: Alberta Children's Hospital, Calgary, Alberta, Canada

	Story booklets		Information sheet	
	N		N	
	70		67	
Age of participant (mean, SD)	32.56	5.31	33.26	6.64
Sex of participant				
Female	54	77.1%	55	82.1%
Male	16	22.9%	12	17.9%
Unknown	0	0.0%	0	0.0%
Sex of child				
Female	26	37.1%	22	32.8%
Male	43	61.4%	45	67.2%
Age of child (median, IQR)	2.34	1.09,4.50	2.09	1.27,3.52
Number adults living in the home				
1	5	7.1%	6	9.0%
2	63	90.0%	47	70.1%
>2	2	2.9%	14	20.9%
Number adults participating in care of child				
1	4	5.7%	4	6.0%
2	60	85.7%	53	79.1%
>2	6	8.6%	10	14.9%
Total number of children living in the home (median, IQR)	2	1,2	2	1,3
Relationship to child				
Parent	68	97.1%	65	97.0%
Other	1	1.4%	1	1.5%
Education				
grades 1-9	1	1.4%	0	0.0%
grades 10-11 (some high school)	5	7.1%	3	4.5%
high school graduate	6	8.6%	11	16.4%
some college/university	14	20.0%	14	20.9%
college graduate	17	24.3%	21	31.3%
post-graduate education or degree	25	35.7%	15	22.4%
Marital status				
never married	4	5.7%	6	9.0%
married/common-law	61	87.1%	55	82.1%
separated, divorced, or widowed	4	5.7%	5	7.5%
Other	0	0.0%	0	0.0%

Household income (Cdn $)				
<15,000	4	5.7%	2	3.0%
15-29,000	3	4.3%	3	4.5%
30-44,000	2	2.9%	4	6.0%
45-59,000	6	8.6%	6	9.0%
60-74,000	6	8.6%	6	9.0%
75-90,000	8	11.4%	10	14.9%
>90,000	33	47.1%	27	40.3%
NR	8	11.4%	9	13.4%
Ethnic or minority group				
No	56	80.0%	44	65.7%
Yes	8	11.4%	18	26.9%
Place of birth				
North America	53	75.7%	50	74.6%
outside of North America	14	20.0%	14	20.9%

SD=standard deviation; IQR=interquartile range

Table G-2. Parental concern at baseline: Demographics: Alberta Children's Hospital, Calgary, Alberta, Canada

	Story booklets		Information sheet	
	N		**N**	
	70		**67**	
Level of concern about the following items:				
uncomfortable aspect of child's cough				
0 (not at all)	3	4.3%	1	1.5%
1	8	11.6%	11	16.4%
2	32	46.4%	30	44.8%
3 (extremely)	26	37.7%	25	37.3%
NR	0	0.0%	0	0.0%
unusual sound or nature of the cough				
0 (not at all)	5	7.1%	1	1.5%
1	11	15.7%	17	25.4%
2	26	37.1%	20	29.9%
3 (extremely)	27	38.6%	29	43.3%
NR	0	0.0%	0	0.0%
unusual sound of child's breathing				
0 (not at all)	5	7.1%	3	4.5%
1	8	11.4%	7	10.4%
2	22	31.4%	26	38.8%
3 (extremely)	35	50.0%	31	46.3%
NR	0	0.0%	0	0.0%
effort that child is making to breathe				
0 (not at all)	6	8.6%	5	7.5%
1	11	15.7%	13	19.4%
2	20	28.6%	20	29.9%
3 (extremely)	33	47.1%	29	43.3%
NR	0	0.0%	0	0.0%
child is not getting enough oxygen				
0 (not at all)	12	17.1%	11	16.4%
1	17	24.3%	12	17.9%
2	25	35.7%	24	35.8%
3 (extremely)	16	22.9%	20	29.9%
NR	0	0.0%	0	0.0%

child may be wheezing or have asthma				
0 (not at all)	17	24.3%	12	17.9%
1	10	14.3%	16	23.9%
2	28	40.0%	18	26.9%
3 (extremely)	14	20.0%	20	29.9%
NR	0	0.0%	1	1.5%
child's sleep was disturbed				
0 (not at all)	9	12.9%	7	10.4%
1	11	15.7%	10	14.9%
2	27	38.6%	20	29.9%
3 (extremely)	23	32.9%	30	44.8%
NR	0	0.0%	0	0.0%
parent felt increasingly tense or frustrated as a result of the illness				
0 (not at all)	17	24.3%	10	14.9%
1	14	20.0%	20	29.9%
2	24	34.3%	22	32.8%
3 (extremely)	15	21.4%	15	22.4%
NR	0	0.0%	67	100.0%
child might be hospitalized				
0 (not at all)	12	17.1%	16	23.9%
1	24	34.3%	30	44.8%
2	21	30.0%	12	17.9%
3 (extremely)	13	18.6%	8	11.9%
NR	0	0.0%	1	1.5%
illness might recur in the future				
0 (not at all)	5	7.1%	9	13.4%
1	22	31.4%	19	28.4%
2	26	37.1%	16	23.9%
3 (extremely)	17	24.3%	23	34.3%
NR	0	0.0%	0	0.0%
not knowing about this illness				
0 (not at all)	13	18.6%	16	23.9%
1	17	24.3%	24	35.8%
2	22	31.4%	14	20.9%
3 (extremely)	16	22.9%	11	16.4%
NR	1	1.4%	2	3.0%
Overall concern (scale 1-10) (mean, SD)	6.10	2.62	6.55	2.33

NR=not reported; SD=standard deviation

Table G-3. History of previous illness, severity of illness at baseline, and ED care: Demographics: Alberta Children's Hospital, Calgary, Alberta, Canada

	Story booklets		Information sheet	
	N		N	
	70		**67**	
History				
Parent first noticed respiratory symptoms (number of days to ED visit) (median, IQR)	1	0,2	1	1,2
prior history of croup				
no history	36	51.4%	27	40.3%
history same child	16	22.9%	15	22.4%
history other child	11	15.7%	11	16.4%
history both	7	10.0%	13	19.4%
prior history of croup admissions				
no admits	60	85.7%	51	76.1%
ED visit only this child	3	4.3%	5	7.5%
ED visit only other child	2	2.9%	4	6.0%
previous admissions this child	4	5.7%	3	4.5%
previous admissions other child	1	1.4%	3	4.5%
prior admissions to ICU				
no ICU admits	69	98.6%	63	94.0%
ICU this child	0	0.0%	1	1.5%
ICU other child	0	0.0%	1	1.5%
prior intubations				
no history	65	92.9%	60	89.6%
history this child	3	4.3%	2	3.0%
history other child	2	2.9%	3	4.5%
history both	0	0.0%	1	1.5%
prior serious illness or chronic medical condition this child				
No	55	78.6%	48	71.6%
Yes	15	21.4%	18	26.9%
Croup severity				
total score (median, IQR)	1		2	
0	21	30.0%	10	14.9%
1	17	24.3%	22	32.8%
2	9	12.9%	14	20.9%
3	10	14.3%	9	13.4%
4	6	8.6%	10	14.9%
5	5	7.1%	2	3.0%
>5	2	2.9%	0	0.0%

missing	0	0.0%	0	0.0%
ED Care				
Disposition				
left without being seen	1	1.4%	4	6.0%
discharged home	63	90.0%	57	85.1%
Admitted	6	8.6%	6	9.0%
Other	0	0.0%	0	0.0%
Prior to recruitment patient seen by				
triage nurse	70	100.0%	67	100.0%
staff nurse	21	30.0%	21	31.3%
Resident	3	4.3%	1	1.5%
staff physician	6	8.6%	5	7.5%
Other	3	4.3%	4	6.0%
Prior to recruitment treatment ordered				
Yes	62	88.6%	58	86.6%
No	5	7.1%	7	10.4%
Read information during ED visit				
Read study material	32	45.7%	45	67.2%
Read additional information	17	24.3%	10	14.9%

ED=emergency department; IQR=inter-quartile range; ICU=intensive care unit;

G-1.2 Primary Outcome: change in parental anxiety from baseline to discharge

The baseline anxiety score on the STAI was 39.2 (SD 12.9) for the story group versus 38.4 (SD 12.1) for the comparison group (Table G-4). At discharge the STAI scores were approximately 7 to 8 points lower for both groups (32.0 and 30.9, respectively). There was no significant difference between groups in change in parental anxiety from baseline to discharge (p=0.83).

G-1.3 Secondary Outcomes

G-1.3.1 Expected future anxiety: The expected future anxiety as measured by the STAI during the Day 1 telephone follow-up showed no significant differences between groups (40.2 versus 39.8, p=0.84). The expected future anxiety was slightly higher than the participants' baseline anxiety (Table G-1).

G-1.3.2 Event Impact: The impact resulting from exposure to anxiety-producing events was measured during the last telephone follow-up; this varied from day 3 to day 9 depending on when symptoms resolved. There were no significant differences between groups either overall (median 7 story group versus 8 comparison group, p=0.672) or for the two subscales: intrusion (median 4.5 for story group versus 3 comparison group, p=0.933) and avoidance (median 1 story group versus 3 comparison group, p=0.445).

G-1.3.3 Parental knowledge: There was no significant difference in knowledge between the two groups during the day 3 follow-up (8.73 versus 8.49, p=0.361). Overall, the knowledge level was high for both groups with a mean of 8.6 (SD 1.46) out of 10.

G-1.3.4 Parental satisfaction: The majority of patients in both groups (64% and 66% respectively) were "very satisfied" with the treatment and care they received in the ED. A further 20% and 27%, respectively, were "somewhat satisfied." The results for satisfaction around their expectations for information were similar with the majority "very satisfied" (86% and 72%) or "somewhat satisfied" (10% and

22%). There was no significant difference between groups in satisfaction with respect to the participants' expectations for treatment and care or their expectations for information.

G-1.3.5 Parental decisional regret: The mean regret score, assessed at 1 day post-ED visit, was not significantly different between groups (1.29 story group [SD 0.510]; 1.20 comparison group [0.316]; p=0.193). None of the five items in the regret scale were significantly different between groups (Table G-5).

G-1.3.6 Incidence of return to be evaluated by a physician (or other health care practitioner) for croup: More participants in the story group returned to a physician or the ED compared to the comparison group; the difference was not significantly different (29.0% versus 30.3%, p=0.867).

G-1.3.7 Healthcare utilization patterns: There were no significant differences between groups in the incidence of contacting a healthcare professional following the ED visit (31.9% story group versus 34.8% comparison group, p=0.715). The most commonly contacted health professional was doctors (21.7% story group versus 24.2% comparison group, p=0.730), followed by return to ED (7.2% story group versus 10.6% comparison group, p=0.493), HealthLink (2.9% story group versus 6.1% comparison group, p=0.373), and other health professional (1.4% story group versus 0% comparison group, p=0.326). The other health professionals contacted was a registered nurse.

G-1.3.8 Resource utilization: No participants used an ambulance following enrollment in the study. No children were hospitalized after being discharged home from the ED. Eight participants in the story group obtained prescription medications after being discharged from the ED compared to 13 participants in the comparison group. Most often prescribed was dexamethasone (n=14), followed by ventolin (n=2), amoxicillin (n=4), zithromax (n=1), prednisone (n=1), Q-var (n=2), and Advair (n=1).

G-1.3.9 Ongoing croup symptoms: Median number of days to no symptoms (TOP score=0) was the same for each group (3 days [IQR 3,5]. The survival distributions for the two groups were significantly different based on the log rank (Mantel-Cox) test (p=0.009) and Tarone-Ware (p=0.028) tests, and of borderline significance based on the Breslow (Generalized Wilcoxon) (p=0.057) (Figure G-2).

Table G-4. Comparison of primary and secondary outcomes: Demographics: Alberta Children's Hospital, Calgary, Alberta, Canada

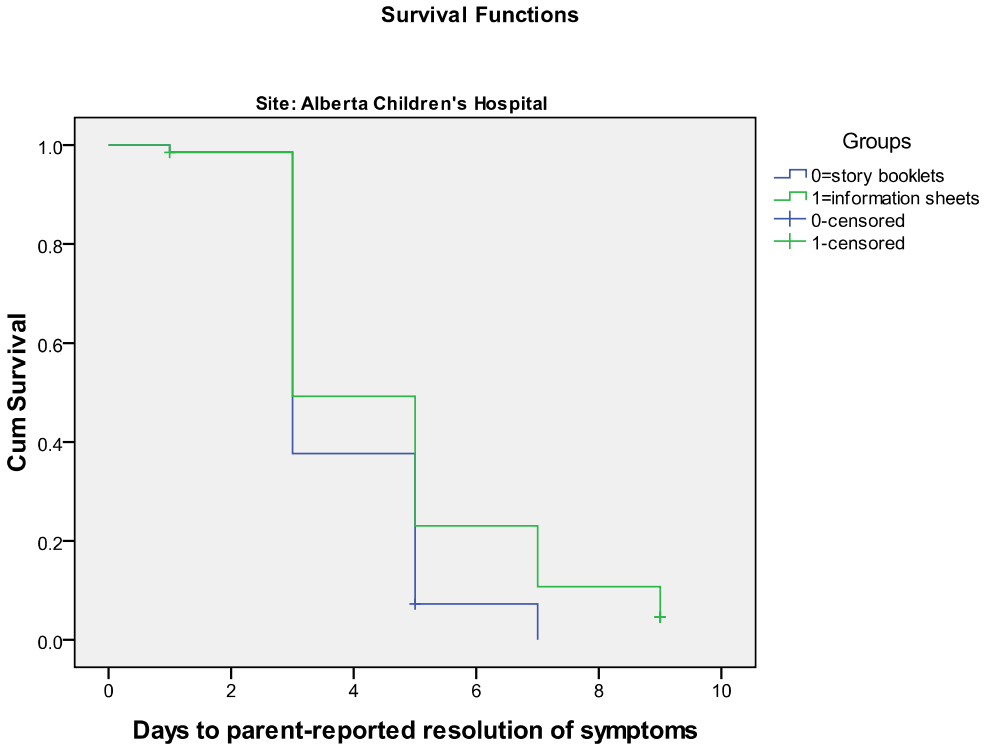
	Story booklets	Information sheet	P-value
	N=70	N=67	
Anxiety - STAI			
Baseline	39.2 (12.9)	38.4 (12.1)	0.699
Discharge	32.0 (11.3)	30.9 (9.32)	0.583
Discharge-Baseline (median, IQR)	8 (2.75, 12)	6 (2, 12)	0.831
Expected anxiety in future episodes of croup (measured at day 1 or 3 post-ED visit) (mean, SD)	40.2 (11.8)	39.8 (11.1)	0.841
Decision Regret (measured at day 1 or 3 post-ED visit)			
mean (SD)	1.294 (0.510)	1.197 (0.316)	0.193
Satisfaction (Day 1 or 3)			
Expectations for treatment and care (n)			0.199
very satisfied	45 (65%)	44 (67%)	
somewhat satisfied	14 (20%)	18 (27%)	
neither satisfied nor dissatisfied	4 (6%)	0	
very dissatisfied	3 (4%)	4 (6%)	
NR	3 (4%)	0	
Expectations for Information (n)			p=0.102
very satisfied	60 (87%)	48 (73%)	
somewhat satisfied	7 (10%)	15 (23%)	
neither satisfied nor dissatisfied	1 (1%)	1 (2%)	
very dissatisfied	0	2 (3%)	
NR	1 (1%)	0	
Knowledge (Day 3) mean (SD)	8.73 (1.59)	8.49 (1.32)	0.361
Impact of event scale (Last follow-up)			
Intrusion sub-scale (median, IQR)	4.5 (1,10.5)	3 (1,10.25)	0.933
Avoidance sub-scale (median, IQR)	1 (0,6.25)	3 (0,6)	0.445
Total (median, IQR)	7 (2,17)	8 (3,16.5)	0.672

STAI=State Trait Anxiety Inventory; SD=standard deviation; ED=emergency department; IQR=inter-quartile range; NR=not reported

Table G-5. Comparison of decision regret scale: Demographics: Alberta Children's Hospital, Calgary, Alberta, Canada

	Story booklets	Information sheet	P-value
	N=69	N=66	
Decision Regret (measured at day 1 or 3 post-ED visit): Parents were asked to respond to the questions regarding their decision to take their child to the ED for the episode of croup in question.			
It was the right decision.			P=0.701
Strongly agree	52	51	
Agree	15	13	
Neither agree nor disagree	0	1	
Strongly disagree	2	1	
I regret the choice that was made.			P=0.563
Strongly agree	2	0	
Agree	0	1	
Neither agree nor disagree	2	2	
Strongly disagree	64	62	
I would go for the same choice if I had to do it over again.			P=0.302
Strongly agree	43	47	
Agree	15	13	
Neither agree nor disagree	3	3	
Strongly disagree	8	2	
The choice did my child a lot of harm.			P=0.322
Strongly agree	0	0	
Agree	2	1	
Neither agree nor disagree	2	0	
Strongly disagree	65	65	
The decision was a wise one.			P=0.383
Strongly agree	53	50	
Agree	14	15	
Neither agree nor disagree	0	1	
Strongly disagree	2	0	

Figure G-2. Comparison of survival functions for time to no symptoms: story booklets (group 0) versus standard information sheet (group 1), Alberta Children's Hospital



G-2. Stollery Children's Hospital

G-2.1 Study sample

Overall 118 parents were recruited at the Stollery Children's Hospital: 59 participants were randomized to receive story booklets and 59 received standard information sheets. Figure G-3 describes the recruitment and follow-up of study participants to day 3 which was the last follow-up point required for all participants.

Characteristics of the trial participants are detailed in Tables G-6 to G-8. There were no notable differences between groups in terms of demographic variables (Table G-6).

Table G-7 presents the results for parental concern at baseline. Overall, parents demonstrated a moderate level of concern with a mean self-rating of 6.24 (SD 2.48) on a scale of 1 to 10, where 10 represents the highest level of concern. The items that generated the most concern were the unusual sound of the child's breathing (47.5% expressed extreme concern) and that the illness might recur in the future (44.1% extreme concern). Other items of concern were the effort the child was making to breathe (39.8% extreme concern) and the unusual sound or nature of the cough (39.8% extreme concern). There appeared to be differences between groups in level of extreme concern, however the pattern was inconsistent across items. For example, with respect to the unusual sound of the child's breathing, 37.3% in the story group expressed extreme concern compared to 57.6% in the comparison group. Conversely, 32.2% in the story group felt extreme increase in tension or frustration as a result of the illness compared to 18.6% in the comparison group.

The majority of participants had no prior history of croup admissions, ICU admissions, or intubations. A substantial proportion (30%) of participants reported a previous experience with croup either with the same or another child, while 22% reported a prior serious illness or medical condition for their child. The most commonly reported serious illnesses/medical conditions was asthma (n=11), heart

problems (n=4), and reflux (n=3). Overall there were no differences between groups in the prevalence of previous medical history or experiences with participants' children.

The majority of patients presented with mild croup with a median croup score of 2 (IQR 0,3) on a scale of 0 to 17. 92% of the patients were discharged home from the ED with only 1 child being admitted (comparison group). Approximately 1 in 3 children had been seen by the staff physician before being recruited into the study. Treatment had already been ordered for 46% of patients prior to recruitment.

Approximately 63% of participants read the study material during their ED stay while 19% read additional information on croup. The groups were similar with respect to reading the study material (61% versus 64%) but more parents in the story group read additional material (23% versus 15%).

Figure G-3. Recruitment and follow-up of study participants

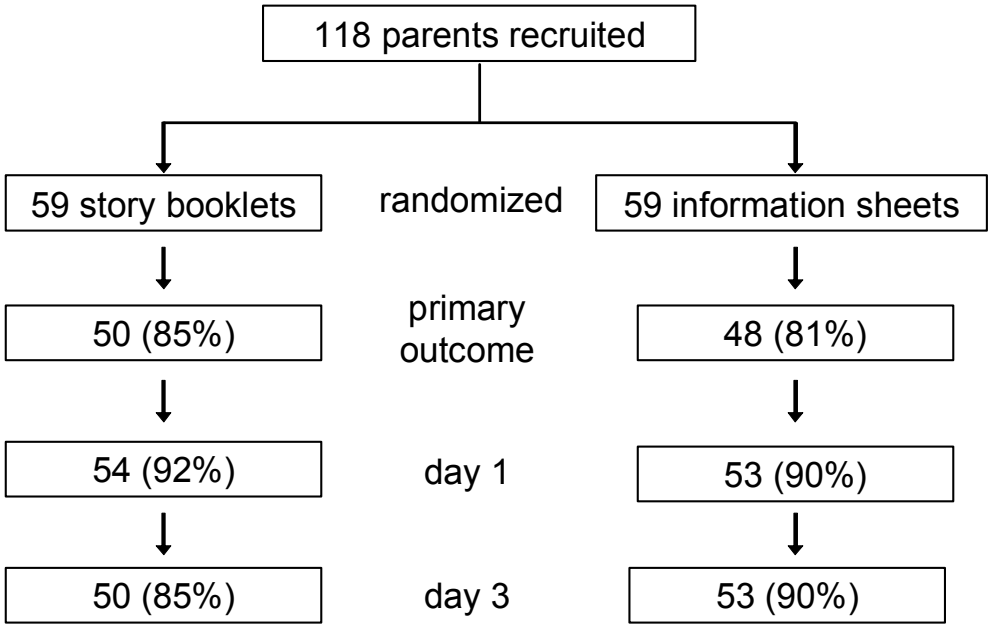


Table G-6. Demographics: Stollery Children's Hospital, Edmonton, Alberta, Canada

	Story booklets		Information sheet	
	N		N	
	59		59	
Age of participant (mean, SD)	33.93	5.81	35.27	6
Sex of participant				
Female	45	76.3%	47	79.7%
Male	13	22.0%	9	15.3%
Unknown	1	1.7%	3	5.1%
Sex of child				
Female	24	40.7%	18	30.5%
Male	35	59.3%	41	69.5%
Age of child (median, IQR)	2.02	1.07,3.11	1.89	1.09,2.93
Number adults living in the home				
1	3	5.1%	5	8.5%
2	45	76.3%	49	83.1%
>2	10	16.9%	5	8.5%
Number adults participating in care of child				
1	1	1.7%	4	6.8%
2	44	74.6%	43	72.9%
>2	13	22.0%	12	20.3%
Total number of children living in the home (median, IQR)	2	1,3	2	1,3
Relationship to child				
Parent	57	96.6%	58	98.3%
Other	1	1.7%	1	1.7%
Education				
grades 1-9	0	0.0%	0	0.0%
grades 10-11 (some high school)	0	0.0%	2	3.4%
high school graduate	14	23.7%	20	33.9%
some college/university	7	11.9%	13	22.0%
college graduate	25	42.4%	18	30.5%
post-graduate education or degree	12	20.3%	6	10.2%
Marital status				
never married	1	1.7%	5	8.5%
married/common-law	53	89.8%	47	79.7%
separated, divorced, or widowed	3	5.1%	6	10.2%

Other	1	1.7%	1	1.7%
Household income				
<15,000	1	1.7%	1	1.7%
15-29,000	2	3.4%	4	6.8%
30-44,000	6	10.2%	4	6.8%
45-59,000	5	8.5%	7	11.9%
60-74,000	8	13.6%	6	10.2%
75-90,000	2	3.4%	9	15.3%
>90,000	28	47.5%	20	33.9%
NR	7	11.9%	8	13.6%
Ethnic or minority group				
No	43	72.9%	48	81.4%
Yes	15	25.4%	10	16.9%
Place of birth				
North America	43	72.9%	51	86.4%
outside of North America	15	25.4%	7	11.9%

SD=standard deviation; IQR=inter-quartile range;

Table G-7. Parental concern at baseline: Stollery Children's Hospital, Edmonton, Alberta, Canada

	Story booklets		Information sheet	
	N		N	
	59		59	
Level of concern about the following items:				
uncomfortable aspect of child's cough				
0 (not at all)	2	3.4%	0	0.0%
1	20	33.9%	11	18.6%
2	20	33.9%	26	44.1%
3 (extremely)	17	28.8%	22	37.3%
NR	0	0.0%	0	0.0%
unusual sound or nature of the cough				
0 (not at all)	3	5.1%	1	1.7%
1	14	23.7%	13	22.0%
2	21	35.6%	19	32.2%
3 (extremely)	21	35.6%	26	44.1%
NR	0	0.0%	0	0.0%
unusual sound of child's breathing				
0 (not at all)	2	3.4%	2	3.4%
1	12	20.3%	8	13.6%
2	23	39.0%	15	25.4%
3 (extremely)	22	37.3%	34	57.6%
NR	0	0.0%	0	0.0%
effort that child is making to breathe				
0 (not at all)	6	10.2%	5	8.5%
1	13	22.0%	12	20.3%
2	18	30.5%	17	28.8%
3 (extremely)	22	37.3%	25	42.4%
NR	0	0.0%	0	0.0%
child is not getting enough oxygen				
0 (not at all)	13	22.0%	10	16.9%
1	15	25.4%	16	27.1%
2	15	25.4%	17	28.8%
3 (extremely)	15	25.4%	16	27.1%
NR	1	1.7%	0	0.0%

child may be wheezing or have asthma				
0 (not at all)	12	20.3%	13	22.0%
1	9	15.3%	14	23.7%
2	18	30.5%	15	25.4%
3 (extremely)	20	33.9%	17	28.8%
NR	0	0.0%	0	0.0%
child's sleep was disturbed				
0 (not at all)	6	10.2%	7	11.9%
1	12	20.3%	10	16.9%
2	16	27.1%	22	37.3%
3 (extremely)	25	42.4%	20	33.9%
NR	0	0.0%	0	0.0%
parent felt increasingly tense or frustrated as a result of the illness				
0 (not at all)	10	16.9%	8	13.6%
1	13	22.0%	12	20.3%
2	17	28.8%	28	47.5%
3 (extremely)	19	32.2%	11	18.6%
NR	0	0.0%	0	0.0%
child might be hospitalized				
0 (not at all)	15	25.4%	12	20.3%
1	13	22.0%	14	23.7%
2	18	30.5%	16	27.1%
3 (extremely)	13	22.0%	17	28.8%
NR	0	0.0%	0	0.0%
illness might recur in the future				
0 (not at all)	5	8.5%	3	5.1%
1	9	15.3%	15	25.4%
2	16	27.1%	18	30.5%
3 (extremely)	29	49.2%	23	39.0%
NR	0	0.0%	0	0.0%
not knowing about this illness				
0 (not at all)	7	11.9%	9	15.3%
1	19	32.2%	17	28.8%
2	14	23.7%	17	28.8%
3 (extremely)	19	32.2%	16	27.1%
NR	0	0.0%	0	0.0%
Overall concern (scale 1-10) (mean, SD)	6.09	2.56	6.39	2.41

NR=not reported; SD=standard deviation

Table G-8. History of previous illness, severity of illness at baseline, and ED care: Stollery Children's Hospital, Edmonton, Alberta, Canada

	Story booklets		Information sheet	
	N		N	
History				
Parent first noticed respiratory symptoms (number of days to ED visit) (median, IQR)	1	0,2	1	1,2
prior history of croup				
no history	33	55.9%	37	62.7%
history same child	10	16.9%	8	13.6%
history other child	8	13.6%	9	15.3%
history both	6	10.2%	5	8.5%
prior history of croup admissions				
no admits	52	88.1%	51	86.4%
ED visit only this child	3	5.1%	2	3.4%
ED visit only other child	0	0.0%	2	3.4%
previous admissions this child	0	0.0%	1	1.7%
previous admissions other child	3	5.1%	3	5.1%
prior admissions to ICU				
no ICU admits	57	96.6%	58	98.3%
ICU this child	0	0.0%	1	1.7%
ICU other child	1	1.7%	0	0.0%
prior intubations				
no history	49	83.1%	50	84.7%
history this child	5	8.5%	5	8.5%
history other child	4	6.8%	3	5.1%
history both	0	0.0%	1	1.7%
prior serious illness or chronic medical condition this child				
No	46	78.0%	45	76.3%
Yes	12	20.3%	14	23.7%
Croup severity				
total score (median, IQR)	1	2		
0	17	28.8%	14	23.7%
1	13	22.0%	9	15.3%
2	9	15.3%	13	22.0%
3	9	15.3%	13	22.0%
4	6	10.2%	5	8.5%
5	2	3.4%	2	3.4%

>5	2	3.4%	3	5.1%
missing	1	1.7%	0	0.0%
ED Care				
Disposition				
left without being seen	0	0.0%	0	0.0%
discharged home	53	89.8%	56	94.9%
Admitted	0	0.0%	1	1.7%
Other	6	10.2%	2	3.4%
Prior to recruitment patient seen by				
triage nurse	53	89.8%	55	93.2%
staff nurse	36	61.0%	37	62.7%
Resident	15	25.4%	22	37.3%
staff physician	22	37.3%	19	32.2%
Other	2	3.4%	2	3.4%
Prior to recruitment treatment ordered				
Yes	25	42.4%	29	49.2%
No	30	50.8%	27	45.8%
Read information during ED visit				
Read study material	36	61.0%	38	64.4%
Read additional information	14	23.7%	9	15.3%

ED=emergency department; IQR=inter-quartile range; ICU=intensive care unit

G-2.2 Primary Outcome: change in parental anxiety from baseline to discharge

The baseline anxiety score on the STAI was 36.1 (SD 11.7) for the story group versus 39.5 (SD 12.3) for the comparison group (Table G-9). At discharge the STAI scores were approximately 4 to 5 points lower for both groups (32.4 and 34.7, respectively. There was no significant difference between groups in change in parental anxiety from baseline to discharge (p=0.47).

G-2.3 Secondary Outcomes

G-2.3.1 Expected future anxiety: The expected future anxiety as measured by the STAI during the Day 1 telephone follow-up showed no significant differences between groups (44.2 versus 46.1, p=0.44). Interestingly, the expected future anxiety was substantially higher than the participants' baseline anxiety (Table G-9).

G-2.3.2 Event Impact: The impact resulting from exposure to anxiety-producing events was measured during the last telephone follow-up; this varied from day 3 to day 9 depending on when symptoms resolved. There were no significant differences between groups either overall (median 7 for story group and 9 for comparison group, p=0.726) or for the two subscales: intrusion (median 4.5 for story group and 4 for comparison group, p=0.298) and avoidance (median 12 for both groups, p=0.773).

G-2.3.3 Parental knowledge: There was no significant difference in knowledge between the two groups during the day 3 follow-up (8.36 versus 8.38, p=0.951). Overall, the knowledge level was high for both groups with a mean of 8.4 (SD 1.42) out of 10.

G-2.3.4 Parental satisfaction: The majority of patients in both groups (64% and 71% respectively) were "very satisfied" with the treatment and care they received in the ED. A further 19% in each group were "somewhat satisfied." The results for satisfaction around their expectations for information were similar with the

majority “very satisfied” (63% and 69%) or “somewhat satisfied” (23% and 20%). There was no significant difference between groups in satisfaction with respect to the participants’ expectations for treatment and care or their expectations for information.

G-2.3.5 Parental decisional regret: The mean regret score, assessed at 1 day post-ED visit, was higher in the story group compared to the comparison group (1.226 versus 1.094). The difference in means was statistically significant (t-test, p=0.016). When the five items in the regret scale were assessed independently, only one item showed a significant difference between groups (Table G-10). More parents in the story group showed less agreement with the statement “I would go for the same choice if I had to do it again” (p=0.039).

G-2.3.6 Incidence of return to be evaluated by a physician (or other health care practitioner) for croup: More participants in the story group returned to a physician or the ED compared to the comparison group; the difference was not significantly different (32.1% versus 18.2%, p=0.096).

G-2.3.7 Healthcare utilization patterns: There was a significant difference between groups in the incidence of contacting a healthcare professional following the ED visit, with more in the story group doing so (34.0% story group versus 17.0% comparison group, p=0.045). The most commonly contacted health professional was doctors (28.3% story group versus 16.4% comparison group, p=0.136), followed by return to ED (9.4% story group versus 1.8% comparison group, p=0.084), HealthLink (7.5% story group versus 0% comparison group, p=0.038), and other health professional (1.9% story group versus 0% comparison group, p=0.306). The other health professionals contacted was a homeopath.

G-2.3.8 Resource utilization: No participants used an ambulance following enrollment in the study. One child in the story group was hospitalized after being discharged home from the ED. Three participants in the story group obtained prescription medications after being discharged from the ED compared to 0

participants in the comparison group. The drugs prescribed were dexamethasone (n=2), motrin (n=1), and ventolin (n=1).

G-2.3.9 Ongoing croup symptoms: Median number of days to no symptoms (TOP score=0) was 3 days (IQR 3,5) for the story group and 5 days for the comparison group (IQR 3,5). The survival distributions for the two groups were not significantly different for any of the statistical tests: log rank (Mantel-Cox), p=0.948; Breslow (Generalized Wilcoxon), p=0.566; and, Tarone-Ware (p=0.713) (Figure G-4).

Table A-9. Comparison of primary and secondary outcomes: Stollery Children's Hospital, Edmonton, Alberta, Canada

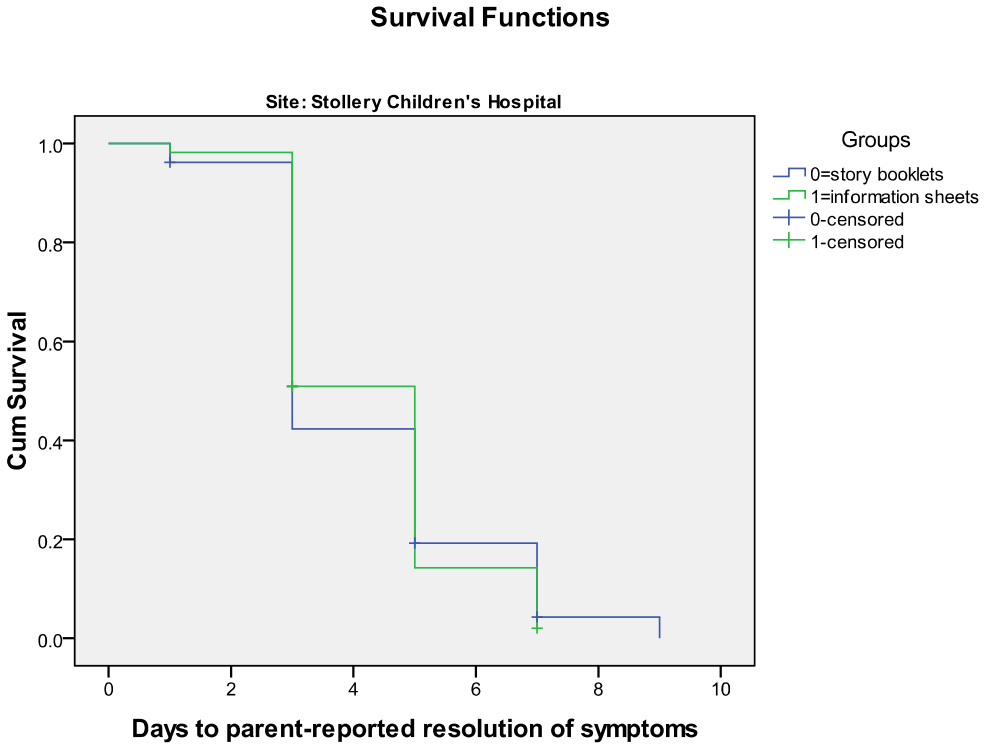
	Story booklets	Information sheet	P-value
	N=59	N=59	
Anxiety - STAI			
Baseline	36.1 (11.7)	39.5 (12.3)	0.128
Discharge	32.4 (10.9)	34.7 (9.85)	0.279
Discharge-Baseline (median, IQR)	4 (1,10.25)	7 (2, 10.25)	p=0.472
Expected anxiety in future episodes of croup (measured at day 1 or 3 post-ED visit) (mean, SD)	44.2 (13.4)	46.1 (12.0)	0.444
Decision Regret (measured at day 1 or 3 post-ED visit)			
mean (SD)	1.226 (0.349)	1.094 (0.182)	0.016
Satisfaction (Day 1 or 3)			
Expectations for treatment and care (n)			P=0.374
very satisfied	38 (72%)	42 (78%)	
somewhat satisfied	11 (21%)	11 (20%)	
neither satisfied nor dissatisfied	0	1 (2%)	
very dissatisfied	2 (4%)	0	
NR	2 (4%)	0	
Expectations for Information (n)			P=0.508
very satisfied	37 (70%)	41 (76%)	
somewhat satisfied	14 (26%)	12 (22%)	
neither satisfied nor dissatisfied	1 (2%)	0	
very dissatisfied	0	1 (2%)	
NR	1 (2%)	0	
Knowledge (Day 3) mean (SD)	8.36 (1.58)	8.38 (1.27)	0.951
Impact of event scale (Last follow-up)			
Intrusion sub-scale (median, IQR)	7 (4,12)	9 (3.75,11.75)	0.726
Avoidance sub-scale (median, IQR)	4.5 (2.75, 9)	4 (0.75,9)	0.298
Total (median, IQR)	12 (7,20)	12 (6.5,21.5)	0.773

STAI=State Trait Anxiety Inventory; IQR=inter-quartile range; ED=emergency department; SD=standard deviation; NR=not reported

Table A-10. Comparison of decision regret scale: Stollery Children's Hospital, Edmonton, Alberta, Canada

	Story booklets	Information sheet	P-value
	N=53	N=54	
Decision Regret (measured at day 1 or 3 post-ED visit): Parents were asked to respond to the questions regarding their decision to take their child to the ED for the episode of croup in question.			
It was the right decision.			P=0.314
Strongly agree	38	46	
Agree	13	7	
Neither agree nor disagree	1	1	
Strongly disagree	1	0	
I regret the choice that was made.			P=0.388
Strongly agree	0	0	
Agree	0	0	
Neither agree nor disagree	4	2	
Strongly disagree	49	52	
I would go for the same choice if I had to do it over again.			P=0.039
Strongly agree	36	48	
Agree	10	4	
Neither agree nor disagree	4	1	
Strongly disagree	3	0	
The choice did my child a lot of harm.			P=0.311
Strongly agree	0	0	
Agree	0	0	
Neither agree nor disagree	1	0	
Strongly disagree	52	54	
The decision was a wise one.			P=0.963
Strongly agree	43	44	
Agree	10	10	
Neither agree nor disagree	0	0	
Strongly disagree	0	0	

Figure G-4. Comparison of survival functions for time to no symptoms: story booklets (group 0) versus standard information sheet (group 1), Stollery Children's Hospital



APPENDIX H. Study proposal to quantify bias in randomized controlled trials in child health

Summary

Context: Bias, or the systematic over or underestimate of a treatment's effect, has important implications for decision making. While the randomized controlled trial (RCT) has been heralded as the gold standard to determine the efficacy of an intervention, it is nonetheless prone to bias. The extent to which bias operates in a given trial can yield inaccuracies of varying magnitude in the estimates of a treatment's effect. The result, at the extremes, is that interventions may be implemented that are not efficacious, or interventions may be withheld that are efficacious. There is a growing body of empirical evidence that quantifies the extent to which different methodological characteristics of a trial exaggerate treatment effects. For example, it has become well recognized that inadequate concealment of allocation and lack of double-blinding can result in overestimates of 18% and 9% respectively, on average.

Rationale: The evidence to date has stemmed from examination of trials involving adult participants. A meta-epidemiological study to quantify bias in a sample of pediatric trials would better inform the design, conduct, and interpretation of research in child health. Further, previous studies have focused on outdated approaches to assessment of "methodological quality." The Risk of Bias tool released in 2008 by The Cochrane Collaboration offers a new paradigm to evaluate methodological characteristics that may be associated with bias. Finally, more research is needed to explore alternative approaches to analysis within meta-epidemiological studies and application to different types of outcomes.

Objectives: The overall goal of the proposed research is to quantify the bias related to specific methodological characteristics in child-relevant RCTs. We will: 1) develop a database of child-relevant systematic reviews of RCTs of therapeutic interventions; 2) describe the RCTs with respect to methodological and study characteristics; and, 3) quantify the association between pre-specified

methodological characteristics and treatment effect estimates and explore variations based on different analytic approaches and types of outcomes.

Design: The sample for this cross-sectional, observational study will be based on systematic reviews in the Cochrane Database of Systematic Reviews. Systematic reviews will be included if they contain at least five trials of children (0 to 17 years of age) that contribute to a meta-analysis.

Main Outcome Measures: Ratios of odds ratio will be generated using logistic regression to compare the treatment effects for trials at high or unclear versus low risk of bias with respect to the following characteristics: sequence generation; allocation concealment; blinding; incomplete outcome data; selective outcome reporting; "other sources of bias;" baseline imbalances; blocked randomization in unblinded trials; early stopping for benefit; influence of trial sponsors; and sample size.

Significance: The result of the proposed work will be empirical evidence of bias associated with various methodological considerations within pediatric trials. This evidence is relevant to a number of stakeholders including researchers, systematic reviewers and meta-analysts, methodologists, and practitioners and other decision-makers. This project will build on the existing evidence base in several ways: 1) it will provide evidence for trials involving an important, vulnerable population where information is currently lacking; 2) it will employ most recent methods for assessing risk of bias in trials; and, 3) it will explore consistency of effects for different statistical approaches and different types of outcome.

H-1. Background

While randomized controlled trials (RCTs) are considered to be the gold standard for evidence on therapeutic interventions,[1] they are nonetheless susceptible to bias.[2] Bias, or the systematic over- or underestimate of a treatment's effect, has important implications for decision making. The implications stem from false positive and false negative results. In practice this may result in the implementation of interventions that are not efficacious and potentially harmful, or withholding of interventions that truly are efficacious. The types of bias that

may occur in RCTs can generally be classified as selection, performance, detection, attrition, and reporting bias.[3] Appendix H-1 provides a description of these different biases. The extent to which these biases operate in a given trial can yield inaccuracies of varying magnitude and direction in the estimates of a treatment's effect.

The internal validity of a study reflects the extent to which the design and conduct of the study have prevented bias.[4] One of the key steps in a systematic review is assessment of a study's internal validity, or potential for bias. With the increase in systematic reviews and development of systematic review methodology over the past 15 years, close attention has been paid to the methods for assessing internal validity. Until recently this has been referred to as "quality assessment" or "assessment of methodological quality."[3] In this context, "quality" refers to "the confidence that the trial design, conduct, and analysis has minimized or avoided biases in its treatment comparisons."[5] To facilitate the assessment of methodological quality, a plethora of tools has emerged.[5-7] These tools often incorporate characteristics that may be associated with bias; however, many tools also contain elements related to reporting (e.g., was the study population described) and design (e.g., was a sample size calculation performed) that are not related to bias.[3]

The Cochrane Collaboration has recently developed a tool to assess the potential risk of bias in RCTs.[3] The Risk of Bias tool is based on six domains: sequence generation, allocation concealment, blinding, incomplete outcome data, selective outcome reporting, and "other sources of bias." Appendix H-1 shows the relationship between these domains and the different types of bias. The Risk of Bias tool was developed to address some of the shortcomings of existing quality assessment instruments. The developers aimed to distinguish between actual methods of conducting the trials versus reporting. Furthermore, the choice of components for inclusion in the tool was based on empirical evidence demonstrating their association with effect estimates. There is a growing body of evidence from methodological studies, and meta-epidemiological studies in

particular, to quantify the extent to which different characteristics of a trial exaggerate treatment effects. Empirical evidence exists for the following characteristics contained within the Risk of Bias tool: sequence generation; allocation concealment; blinding; incomplete outcome reporting; selective outcome reporting; trials stopped early for benefit; and, inappropriate influence of the funder (Appendix H-2).

Appropriate methods for generating the randomization sequence and concealing the allocation sequence are essential to minimize selection bias. Randomization ensures that the groups being compared are balanced with respect to both known and unknown confounders, while allocation concealment ensures that the randomization sequence is unknown to the person entering participants into a trial until allocation to an intervention group has occurred. Four studies have evaluated the association between adequate and inadequate sequence generation and effect estimates based on meta-analyses in a variety of clinical areas (Table H2-A).[8-11] Pooled results from these four meta-epidemiological studies indicate that inadequate sequence generation results in overestimation of treatment effects by 12% (ROR 0.88, 95% CI 0.79-0.99).[12] Seven published studies have independently examined the association between allocation concealment and treatment effect estimates.[8-11;13-15] Pooled results from these studies show that studies with inadequate allocation concealment exaggerate treatment effects by 18% (ROR 0.82, 95% CI 0.71-0.94).[15] The effect is not consistent across different types of outcomes: the effect has been found to be less, and not significant, for all-cause mortality; while significant and heterogeneous for other outcomes.[16;17] Preliminary evidence suggests some variability in effect by degree of between-trial heterogeneity.[18]

"Double-blinding" has long been considered a methodological characteristic of importance.[19] Blinding of key individuals in a trial (i.e., study participants, study personnel, and outcome assessors) can minimize performance and detection bias. Seven studies have independently evaluated the impact of double-blinding on effect estimates.[8-11;13-15] Pooled results from these studies show a 9% overestimate

for studies that were not described as double-blind; the result is of borderline significance (ROR 0.91, 95% CI 0.83,1.0).[15] More recent preliminary evidence suggests that blinding may not be significantly associated with effect estimates (ROR 0.97, 95% CI 0.92,1.01); however, it is not clear how blinding was defined in this study.[17] A limitation of the majority of previous studies is that they rely solely on reporting of "double-blinding."[20;21] One study examined blinding of patients, caregivers, and outcome assessors separately and found no consistent trends in treatment effects.[13] More recently, experts maintain that it is more important to look at who is blinded in a trial [21] and the consequences of inadequate blinding [22].

The effect of missing outcome data and how missing data are managed has been investigated in a number of studies. Several studies have suggested that per protocol analyses may yield more favorable treatment estimates compared to intention-to-treat (ITT) analyses;[23-25] these effects may be exaggerated at the meta-analysis level.[23] A recent study found that "modified" ITT versus ITT analyses exaggerated effect estimates by 15% (ROR 0.85, 95% CI 0.81,0.88).[26] However, four meta-epidemiological studies have provided no evidence to suggest that missing outcome data are associated with effect size estimates. Each study examined the issue in a different way. Schulz et al. found no difference in effect estimates for studies that reported exclusions (ROR 1.07, 95% CI 0.94,1.21).[8] Kjaergard and colleagues found no difference for studies that reported the number and reasons for exclusions (ROR 1.50, 95% CI 0.80,2.78).[10] Balk et al. assessed four dimensions (drop-outs recorded, reasons for drop-outs given, percentage of drop-outs, intention-to-treat analysis) and found no significant associations with effect estimates (Appendix H-2, Table H2-D).[13] Finally, Siersma et al. found no association between studies that performed ITT and effect estimates (ROR 1.01, 95% CI 0.93,1.11).[11] A recent meta-epidemiological study found no significant difference in effect sizes overall for studies with adequate (i.e., ITT) versus inadequate or unclear approaches to analysis (ES -0.09, 95% CI -0.23,0.05), but results varied according to the degree of between-trial heterogeneity.[27]

Empirical investigations of selective outcome reporting have emerged more recently. Selective outcome reporting occurs within-studies and is defined as "the selection of a subset of the original variables recorded for inclusion in publication of trials."[28] The most apparent source of bias is when outcomes measured in a trial are not reported based on their statistical significance; however, other sources of selective outcome reporting exist, such as how the outcome is analyzed, how and when the outcome is measured, as well as reporting of different subsets of data or subgroups.[29-32] A recent systematic review summarized five studies that followed inception cohorts from protocol to full publication in order to examine selective reporting of outcomes.[29;33-37] Four studies "that examined the association between outcome reporting bias and statistical significance found that statistically significant outcomes were more likely to be completely reported than non-significant outcomes (range of odds ratios: 2.2 to 4.7)."[29] The studies also found discrepancies in the primary outcomes proposed and those reported. Furukawa et al. examined the impact of selective outcome reporting on the results of meta-analysis.[38] They found that approximately half of the trials identified as relevant to a systematic review did not contribute to the meta-analysis of patient-important outcomes, and the effect estimates decreased as the proportion of relevant studies contributing to the meta-analysis increased. Other research has investigated discrepancies due to unpublished versus published scales [32] and handling of baseline and endpoint data [39].

The final domain within the Cochrane tool refers to "other sources of bias." This represents an assortment of study characteristics that may lead to biased results, including factors associated with specific designs (e.g., cross-over trials, cluster trials). The characteristics within this domain that are relevant to this proposal include: early stopping for benefit; inappropriate influence of study sponsor; blocked randomization in unblinded trials; and, baseline imbalances. A systematic review of trials stopped early for benefit provides empirical evidence that such trials overestimate treatment effects; effect estimates vary by number of events, with exaggerated estimates more pronounced in trials with fewer endpoints.[40] The spurious results of trials stopped early for benefit can extend to meta-analysis,

where their impact may be substantial.[41] Reports of inappropriate influence of funders in terms of reporting and publication abound. Evidence shows that published research that is industry-sponsored is more likely to have results or conclusions favouring the sponsor.[42-44] Further, evidence based on trial protocols shows that industry-sponsors often have access to data during the conduct of a trial and authority to stop the trial at any point or prevent publication of trial results.[45] Bias related to blocked randomization in unblinded trials and baseline imbalances is supported by theoretical principles.[1;46] Sample size is not included in the Risk of Bias tool; however, some evidence suggests that small samples may be associated with exaggerated effect estimates.[10;47] This variable warrants investigation within child health research given the preponderance of trials with small samples.[48;49]

H-2. Rationale

The existing evidence has begun to quantify different biases in randomized trials; however, there are some inconsistencies across studies and clinical areas. Moreover, "the evidence base remains incomplete."[3] The evidence to date has stemmed primarily from examination of trials involving adult participants. No meta-epidemiological studies have focused specifically on pediatric trials. Further, those that have included some pediatric trials in their samples have addressed these as a homogeneous group. There is recognition that biases may vary across different clinical areas and investigation within different areas is warranted.[13;14] In fact, Balk et al. found variation in the direction of effects across studies which "calls into question whether any of these associations could provide a general rule for evaluating RCTs across clinical areas." A meta-epidemiological study to quantify bias in a sample of pediatric trials would better inform the design, conduct, and interpretation of research in child health.

There are many ways that "quality measures" are defined.[13] The inconsistency in applying and defining "quality" criteria has been cited as a limitation of previous research.[15] In the vast majority of previous meta-epidemiological research,

"quality measures" have been based on reporting. For instance, the majority of studies that have evaluated blinding assessed whether or not the study was described as double-blind. As mentioned above, the different ways in which missing outcome data have been assessed illustrates the various approaches to evaluating this variable and illuminates potential areas for improvement in subsequent research. The approach in this project will be cutting-edge, in that domains will be evaluated based on "risk of bias" rather than reporting. We will employ the new Cochrane Risk of Bias tool which represents a new paradigm, based on empirical evidence, for evaluating study characteristics that may be associated with bias.

Much of the accumulating evidence for bias in trials is based on meta-epidemiological studies. This approach is advocated as it minimizes confounding due to disease and interventions.[3] The majority of meta-epidemiological studies have followed the same general approach, wherein logistic regression is used to generate an overall estimate of the ratio of odds ratios from individual studies while controlling for the effects of treatment, trial, meta-analysis, and other co-variates of interest.[2] Other approaches have been proposed to overcome some of the limitations of the logistic regression analysis, in particular the assumption of homogeneity of bias across trials and across meta-analyses.[2;11] There is some evidence that the different statistical approaches may yield different results.[50] The majority of meta-epidemiological studies have focused on binary outcomes which may limit the generalizability of study findings. More recent meta-epidemiological research provides some models for use of continuous outcomes.[18;27;51] Finally, results may vary for different summary measures (e.g., odds ratios, relative risks, risk differences). This study will explore different statistical methods within meta-epidemiological studies. Specifically we will explore the two more common approaches to statistical analysis in meta-epidemiological studies [2] and the evaluation of continuous in addition to binary outcomes.

H-3. Goals and Objectives

The overall goal of this project is to quantify the extent of bias related to specific methodological characteristics in child-relevant RCTs.

The specific objectives are:

1) To develop a database of child-relevant systematic reviews of RCTs of therapeutic interventions;
2) To describe the RCTs with respect to methodological and study characteristics;
3) To quantify the association between pre-specified methodological characteristics and treatment effect estimates and to explore variations based on analytic approach and type of outcome.

H-4. Hypothesis

The null hypothesis is that there will be no difference in treatment effect estimates for trials at high or unclear versus low risk of bias with respect to: sequence generation; allocation concealment; blinding; incomplete outcome data; selective outcome reporting; "other sources of bias;" early stopping for benefit; influence of trial sponsors; baseline imbalance; blocking in unblinded trials; and sample size.

The alternative hypothesis is that treatment effect estimates will be significantly different for the same comparisons (i.e., effect estimates will be different in trials that are at high or unclear versus low risk with respect to the given methodological characteristic).

H-5. Study Protocol

H-5.1 Study Design

This will be an observational study based on a sample of RCTs contributing to the meta-analyses identified within systematic reviews. The study is cross-sectional in that the trials have already been conducted, hence the study variables and

outcomes have already occurred and data will be collected on all variables and outcomes concurrently.

H-5.2 Study Sample

The study will be based on systematic reviews relevant to child health. The sampling frame will be the Cochrane Database of Systematic Reviews (CDSR). A research librarian with the Cochrane Child Health Field completed a search of the CDSR for child-relevant systematic reviews in October 2008 (see Appendix H-3 for search strategy). The search yielded 1,593 titles, of which 877 were completed reviews that could be considered for eligibility in the present study. Appendix H-4 presents a QUOROM diagram for initial screening of the reviews.

The CDSR was chosen for the sampling frame for the following reasons: 1) Cochrane reviews provide tabulated data from the component trials as well as detailed descriptions of key characteristics (e.g., study population); 2) Cochrane reviews provide a detailed list of references for all relevant trials; 3) Cochrane reviews have been reported to be of higher quality which may translate into more comprehensive searches, hence more variability with respect to methodological characteristics; 4) the CDSR offers a more homogeneous sample with respect to domains (i.e., therapeutic effectiveness) and restricted to RCTs.

Assessing eligibility for the sample will occur at three levels: content area; systematic review; and, RCT. The sample of systematic reviews will be based on content areas. The clinical areas of the potentially eligible reviews are listed in Appendix H-4. The clinical areas are based on Cochrane Collaborative Review Groups. Details regarding the scope of each group are available at each group's website, through http://www.cochrane.org/contact/entities.htm#CRGLIST. We will screen reviews for eligibility beginning with the groups with the largest number of reviews. The assumption is that this may reflect the content of trials in child health in general, and indirectly the priority areas for child health (i.e., neonatology; airways; acute respiratory infections; developmental, psychosocial

and learning problems; infectious diseases).[52] Reviews will be screened until the required sample size of trials is met (see Section 5.4.2 Sample Size).

Systematic reviews will be included based on the following criteria:

a) Systematic reviews must have a minimum of five RCTs[14;53] involving only pediatric patients (ages 0 to 17 years), and a maximum of 40 RCTs,[15] that contribute to at least one meta-analysis.
b) The systematic review must address a question of therapeutic effectiveness, and may include outcomes of efficacy, harm, or both.

Systematic reviews must include five RCTs that meet the following criteria:

a) Trials are described as, or claim to be, randomized.[36] RCTs are operationally defined as "a prospective study assessing the efficacy or harm of health care interventions and randomly allocating human participants to study groups."[34]
b) Trials must be superiority studies with parallel designs involving at least 2 comparison groups.
c) Reports of trials must be "full-length"[10;11] and language of publication must be English, French, Spanish, or German in order to facilitate risk of bias assessments.[8;9] Where two reports exist for the same trial, the most comprehensive report will be used for risk of bias assessments and data extraction.
d) Duplicate trials will be removed. A random numbers table will be used to determine from which meta-analysis the duplicate trial will be removed,[8] with provisions to ensure that each meta-analysis has at least five trials.

H-5.3 Data Extraction

Table H-1 provides a summary of variables and data to be extracted. These are summarized by study outcomes (H-5.3.1); study characteristics (H-5.3.2); and, methodological characteristics (H-5.3.3).

H-5.3.1 Study Outcomes: A matrix will be developed for each SR listing the outcomes that were meta-analyzed and the studies contributing to each.[3] When

possible, one binary and one continuous outcome will be selected from each systematic review based on the largest number of trials contributing data for that endpoint to a meta-analysis [9;13] (where at least five RCTs and fewer than 40 RCTs [15] have contributed to the meta-analysis). For binary outcomes, the numbers in each group with or without the event and the total number in each group will be identified. For continuous outcomes, the mean and standard deviation for each group will be identified. The data in the systematic review will be checked against the primary report for each study. For each meta-analysis we will document the methods used to pool results (i.e., summary measure and model). The outcomes will be categorized as efficacy or harm and objective or subjective based on previously reported criteria (Appendix H-5). Further, whether or not a validated (and/or published) outcome measurement tool was used will be documented.

H-5.3.2 Study Characteristics: In addition to the methodological characteristics of interest, the following study characteristics will be extracted for each trial: year of publication; publication status; single versus multi-center; type of intervention;[11] type of control;[13] blinding of participants and/or parents;[54] blinding of investigators; blinding of outcome assessors; completeness of outcome reporting;[35] and, source of funding (See Appendix H-5 for classification of variables).

H-5.3.3 Methodological Characteristics: The following methodological characteristics will be assessed for each trial: sequence generation; allocation concealment; blinding; incomplete outcome reporting; selective outcome reporting; "other sources of bias;" baseline imbalance; trials stopped early for benefit; blocked randomization in unblinded trials; inappropriate influence of trial sponsors; and, sample size. The choice of methodological characteristics was driven by the empirical evidence that exists confirming or suggesting an association with biased estimates of treatment effect (Appendix H-2). Each methodological characteristic will be assessed as high, unclear, or low risk of bias based on guidelines that accompany the "risk of bias" tool (Appendix H-6 for Summary Table),[3] with specific modifications. For selective outcome reporting,

we will compare the presented results with the outcomes mentioned in the methods section of the same article.[35;36] Current research by our group has shown that it is difficult to access protocols for trials. Among a random sample of trials published in 2007, protocols were available for only 19 of 85 studies (22%). Further, previous research has found that the protocols often do not contain sufficient detail for assessments to be made.[33] Sample size will be categorized as large (low risk; minimum 200 patients across two groups [1;47]) and small (high risk; less than 200 patients).

H-5.3.4 Methods: A data extraction form and instructions will be developed to capture study characteristics, methodological characteristics (i.e., risk of bias), and outcome data. The data extraction form will be pilot tested by all members of the study team (co-investigators and study personnel) using five trials, from the clinical areas represented in the final sample of systematic reviews, that are selectively chosen to reflect a range in terms of risk of bias.[3] The co-investigators will represent individuals with both clinical and methodological expertise. Revisions to the data extraction form and accompanying instructions will be made based on discrepancies, uncertainties, and ensuing discussions. Another five trials will be assessed by the same individuals based on the revised form. Further revisions will be made as necessary to ensure clarity and consistency. Inter-rater reliability will be assessed for each pilot phase using weighted kappa.[55] Subsequent pilot testing will occur if the inter-rater reliability (kappa) is less than 0.61.[9;56] Two individuals will independently extract data from each trial. Discrepancies will be resolved through discussion and referring to the original report; where discrepancies cannot be resolved between the pair of data extractors, a third person (co-investigator) will adjudicate. Inter-rater agreement will be assessed using weighted kappa.[56]

H-5.4 Statistical Considerations

H-5.4.1 Descriptive Analysis: The study sample will be described in terms of the study characteristics and methodological characteristics listed above (Sections 5.3.2 and 5.3.3). Frequencies and percentages will be used for this purpose.

H-5.4.2 Analysis of Bias: The primary analysis will be based on binary outcomes analyzed using logistic regression models described below. Secondary analyses will be based on the meta-meta-analysis detailed below. The second approach will also be applied to the continuous outcomes.

Logistic regression. Endpoints will be recoded so that the outcome occurrence is undesired (i.e., death rather than survival); hence, an odds ratio of less than one suggests that the treatment is beneficial. For each trial, we will calculate a log odds ratio and standard error of the odds ratio for the effect of treatment on the binary outcome of interest.[11] We will develop a logistic regression model with indicator variables for the effects of treatment, the interaction between treatment and methodological characteristic, and the interaction between treatment and meta-analysis.[2] Only meta-analyses containing at least one high or unclear and one low risk study with respect to a given methodological characteristic will be included in the analysis for that characteristic.[14] The coefficient for the treatment/methodological characteristic interaction term provides a log of the ratio of odds ratio. The ratio of odds ratio is a measure of the odds ratio for high or unclear risk trials relative to the odds ratio for low risk trials for the given methodological characteristic, and can be interpreted as the percent of exaggeration in effect estimates for trials at high or unclear versus low risk of bias. A ratio of odds ratio less than one suggests that the treatment effect in the comparison category (i.e., high or unclear risk) is greater than in the reference category (i.e., low risk). We will calculate a 95% confidence interval using robust standard errors [14;16] for each ratio of odds ratio estimate. We will present a p-value for the test of interaction between treatment and methodological characteristic. Studies with high and unclear risk will be combined if the results of effect estimates are similar in terms of direction, magnitude, and overlap of confidence intervals, or if there are insufficient numbers in either category to yield a stable model. Methodological characteristics that demonstrate a significant difference will be further explored in purposeful models to assess the effects simultaneously. We will use approximate *F* ratio tests to assess between-trial heterogeneity based on the mean residual deviance of the fitted models.[8;9] Finally, we will explore

whether effects of the methodological characteristics vary for different study characteristics including: publication status (published versus unpublished); source of funding (industry versus non-industry); nature of the outcome (efficacy versus harm; objective versus subjective); type of intervention (drug versus non-drug); and, type of control (active versus inactive; placebo versus other).

Meta-meta-analysis. As above, endpoints will be coded so that the outcome occurrence is undesired. Within each meta-analysis, we will generate a ratio of pooled estimates. We will use the summary statistic and model that was used in the original meta-analysis. The ratios for each meta-analysis will be combined using meta-analytic techniques with inverse-variance weighting and a random effects model.[2] Between meta-analysis heterogeneity will be described using the I^2 statistic and assessed using the chi-squared test.[3] We will use the same approach stratifying studies by variables other than meta-analysis (i.e., publication status, source of funding, nature of outcome, type of intervention, type of control), without pooling across sub-groups, in an effort to examine biases within homogeneous subsets.[11] A similar approach will be applied to the continuous outcomes using mean differences.

Analyses will be performed using Review Manager version 5.0 (Nordic Cochrane Centre, The Cochrane Collaboration, Copenhagen) and Stata version 7.0 (Stata Corporation, College Station, Texas). Significance level will be set at p=0.10 for tests of heterogeneity and p=0.05 otherwise.

H-5.4.2 Sample Size: There are few precedents in the literature for calculating sample sizes in meta-epidemiological studies.[38] In fact, two previous methodological studies based their sample size on anticipated workload [15] and time constraints [32]. Sample size for another study was based on the sample size used in a previous similar study.[10] We will also take a pragmatic approach to determine sample size. The largest meta-epidemiological study to date, exclusive of meta-meta-epidemiological research, had a sample size of 523 trials from 41 systematic reviews.[11] We will plan for a sample size of 500 trials from a number

of systematic reviews that is proportionate to the number of potentially eligible systematic reviews in the most frequently represented clinical areas.

Table H-1. Study variables: categorization and use in analysis

Characteristic/Outcome	Categorization	Analysis
Study Characteristics		
year of publication	Year	descriptive
publication status	published/unpublished (Appendix H-5)	effect modifier
multi-center versus single-center	multi/single-center	descriptive
type of intervention	Appendix H-5	descriptive
	drug/non-drug	effect modifier
nature of intervention	efficacy/harm	effect modifier
	objective/subjective (Appendix H-5)	effect modifier
type of control	Appendix H-5	descriptive
	active/inactive	effect modifier
blinding of participants	yes/no	descriptive
blinding of parents	yes/no	descriptive
blinding of investigators	yes/no	descriptive
blinding of outcome assessors	yes/no	descriptive
blinding, others	yes/no	descriptive
completeness of outcome reporting	Appendix H-5	descriptive
source of funding	Appendix H-5	descriptive
	industry/non-industry	effect modifier
Methodological Characteristics/ Risk of Bias		
sequence generation	high/low/unclear risk	independent variable
allocation concealment	high/low/unclear risk	independent variable
blinding	high/low/unclear risk	independent variable
missing outcome	high/low/unclear risk	independent variable
selective outcome reporting	high/low/unclear risk	independent variable
"other sources of bias"	high/low/unclear risk	independent variable
trial stopped early for benefit	high/low/unclear risk	independent variable
baseline imbalance	high/low/unclear risk	independent variable
influence of trial sponsor	high/low/unclear risk	independent variable
block randomization in unblinded trials	high/low/unclear risk	independent variable
sample size	high/low/unclear risk	independent variable
Outcomes		
binary outcomes	number of events and number of individuals in treatment and control groups for each trial	dependent variable
continuous outcomes	mean, standard deviation and sample size for treatment and control groups in each trial	dependent variable
Analysis	summary measure and model used to pool results for each meta-analysis	analytic approach

H-6. Future Research

There is a need for future research in three different areas: 1) bias in trials in general, and specifically in child health; 2) validity of risk of bias assessments; and, 3) statistical approaches for meta-epidemiological research.

The majority of meta-epidemiological research on bias has investigated sources of bias independently.[11] The relative importance and interactions of different biases, as well as the influence of other study factors (e.g., source of funding), warrants closer attention.[14] For example, Dwan et al. recommended empirical evaluations of both outcome reporting bias and study publication bias to understand their relative importance.[29] Further work is also needed to elucidate the different sources of bias and the effects of bias in studies of various designs, including cross-over, cluster, equivalence, and non-inferiority.

Two methodological studies of bias merit replication within the child health context. First, the investigation of outcome reporting bias in an inception cohort of protocols would provide empirical evidence specific to child health. A cohort of sufficient size would be required. This could potentially be identified through a large institution specific to children (e.g., The Hospital for Sick Children in Toronto) or across several similar institutions. Second, the effect of outcome reporting bias in terms of meta-analysis would be valuable for systematic reviewers and users of systematic reviews. One study to date has assessed the change in effect estimates based on the proportion of relevant trials contributing data.[38] This could be readily replicated using a sample of meta-analyses taken from the database of systematic reviews that will be developed as part of this study.

A limitation of previous meta-epidemiological research on bias is the variation with which methodological characteristics are defined and categorized. We have sought to improve on this by relying on risk of bias assessments defined by the Cochrane tool. However, limitations persist in that judgments continue to be made

based on reporting and not necessarily conduct.[3] Further research to validate risk of bias assessments and the Cochrane tool is needed.

The majority of meta-epidemiological research to date has employed similar methods. Leaders in this field have recognized that "too little consideration has so far been given to appropriate statistical methods for this type of meta-epidemiological research."[14] One study has explored different modeling techniques [11] and another study used a Bayesian approach [13]. The former study proposed various models to address some of the assumptions that are made in the more accepted approaches, specifically homogeneity across trials and meta-analyses. A recent letter in the *Annals of Internal Medicine* presented corrected estimates of previously published results based on a different statistical analysis that allowed for stratification by meta-analysis.[10;50] Further research in this area could enhance the accuracy and generalizability of results stemming from this type of work.

H-7. Significance of the Proposed Work

The result of the proposed work will be empirical evidence of bias associated with various methodological considerations within pediatric trials. This evidence is relevant to a number of stakeholders including: 1) researchers (when designing and executing future trials); 2) systematic reviewers and meta-analysts (when undertaking "quality assessment" and interpreting the results of a systematic review); 3) methodologists (when designing and implementing approaches to "quality assessment" or risk of bias in trials); and, 4) practitioners and other decision-makers (when interpreting data from clinical trials and making decisions that impact patient care). This project will build on the existing evidence base in several ways: 1) it will provide evidence for trials involving an important, vulnerable population where information is currently lacking; 2) it will employ most recent methods for assessing risk of bias; and, 3) it will explore consistency of effects for different statistical approaches and across different types of outcomes.

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APPENDIX H-1

A COMMON CLASSIFICATION SCHEME FOR BIAS

Type of bias	Description	Relevant domains in the Collaboration's 'Risk of bias' tool
Selection bias.	Systematic differences between baseline characteristics of the groups that are compared.	• Sequence generation; • Allocation concealment.
Performance bias.	Systematic differences between groups in the care that is provided, or in exposure to factors other than the interventions of interest.	• Blinding of participants, personnel and outcome assessors; • Other potential threats to validity.
Attrition bias.	Systematic differences between groups in withdrawals from a study.	• Incomplete outcome data; • Blinding of participants, personnel and outcome assessors.
Detection bias.	Systematic differences between groups in how outcomes are determined.	• Blinding of participants, personnel and outcome assessors; • Other potential threats to validity.
Reporting bias.	Systematic differences between reported and unreported findings.	• Selective outcome reporting; • (see also Chapter 10).

From: Higgins JPT, Green S (editors). Cochrane Handbook for Systematic Reviews of Interventions *Version 5.0.1 (Table 8.4.a) [updated September 2008]. The Cochrane Collaboration, 2008. Available from www.cochrane-handbook.org.*

APPENDIX H-2

SUMMARY OF EMPIRICAL EVIDENCE FOR BIAS

TABLE H2-A. EMPIRICAL EVIDENCE FOR SEQUENCE GENERATION

Study	Content Area	Sample Size	Results (95% CI)	Comments
Published Studies				
Schulz 1995[8]	pregnancy and childbirth	33 MAs with 250 trials (62,091 participants; 12,030 outcome events)	ROR 0.95 (0.81,1.12), n=229 trials; controlling for AC, exclusions, DB	Sequence generation may be important in trials with adequate AC (ROR 0.75 [0.55,1.02], n=79 trials).
Moher 1998[9]	digestive, circulatory, mental health and pregnancy and childbirth	11 MAs with 127 trials (10,492 patients)	ROR 0.89 (0.67,1.2)	Quality assessments performed blinded.
Kjaergard 2001[10]	8 therapeutic areas: cardiology, surgery, pregnancy, schizophrenia, gynaecology, addictions, hypertension, neonatal	14 MAs with 190 RCTs (136,164 participants)	ROR 0.49 (0.30,0.81)*	Also compared large trials versus small trials with varying methodological quality, as well as small trials with varying methodological quality.
Balk 2002[13]	4 medical areas: cardiovascular, infectious disease, pediatrics, surgery	26 MAs with 276 trials	ROR 1.03 (0.89,1.02)	Authors conclude that quality is only one potential explanation for heterogeneity in treatment effect and should not be over-interpreted.
Siersma 2007[11]	MAs randomly selected from CL	523 trials from 41 reviews with 48 meta-analyses	ROR 0.84 (0.78,0.91)†	Only variable consistently found to be significant across different statistical methods/models was sequence generation.

Unpublished Studies				
Als-Nielsen 2004[12]	review of 5 empirical studies	not specified	ROR 0.88 (0.79,0.99)	Considerable heterogeneity among studies; impact of bias seems to vary considerably across interventions and disease areas.
Savovic 2008[17]	review of previous studies (number unclear)	119 MAs with 1,038 trials	ROR 0.87 (0.83,0.92)	Significant between MA heterogeneity. Bias less for all-cause mortality versus other outcomes.

CI=confidence interval; MA=meta-analysis; ROR=ratio of odds ratio; AC=allocation concealment; DB=double-blinding; RCT=randomized controlled trial; CL=Cochrane Library

* Revised analyses published in 2008 showed no statistically significant association for sequence generation (ROR 0.95 [0.86,1.04]).[50]

† Results vary by statistical model: results presented are based on logistic regression; however, results are not significant when stratification approach is used, as was seen for Kjaergard 2001*

TABLE H2-B. EMPIRICAL EVIDENCE FOR ALLOCATION CONCEALMENT

Study	Sample	Size	Results (95% CI)	Comments
Published Studies				
Schulz 1995[8]	pregnancy and childbirth	33 MAs with 250 trials (62,091 participants; 12,030 outcome events)	unclear AC: ROR 0.67 (0.6,0.75); inadequate AC ROR 0.59 (0.48,0.73) (unadjusted for other quality measures) (n=250); unclear AC: ROR 0.70 (0.62,0.79) (adjusted for other SG, DB, withdrawals) (n=229)	Inadequate AC may be a surrogate for other quality measures so magnitude of associations may reflect biases other than selection biases; estimates for unclear AC were heterogeneous across meta-analyses
Moher 1998[9]	digestive, circulatory, mental health and pregnancy and childbirth	11 MAs with 127 trials (10,492 patients)	ROR 0.63 (0.45,0.88)	Majority of outcomes were objective. Quality assessments performed blinded.
Kjaergard 2001[10]	8 therapeutic areas: cardiology, surgery, pregnancy, schizophrenia, gynaecology, addictions, hypertension, neonatal	14 MAs with 190 RCTs (136,164 participants)	ROR 0.60 (0.31,1.15)*	Also compared large trials versus small trials with varying methodological quality, as well as small trials with varying methodological quality.
Balk 2002[13]	4 medical areas: cardiovascular, infectious disease, pediatrics, surgery	26 MAs with 276 trials	relative OR 1.05 (0.91,1.21)	quality is only one potential explanation for heterogeneity in treatment effect and shouldn't be over-interpreted
Egger 2003[14]	MAs in four disease areas: infectious diseases, neurology, obstetrics/gynaecology, other/miscellaneous	39 MAs with 304 trials	ratio of pooled estimates 0.79 (0.7,0.89)	Significant heterogeneity between MAs. Effect more pronounced for active control interventions (chance finding?).

	pooled results of 4 empirical studies (Schulz, Moher, Kjaergard, Egger)		ratio of effect estimates 0.71 (0.66,0.77)	
Siersma 2007[11]	MAs randomly selected from CL	523 trials from 41 reviews with 48 meta-analyses	ROR 1.01 (0.94,1.10)	Results consistent in terms of statistical significance for different statistical approaches.
Pildal 2007[15]	randomly selected MAs from PubMed and CL	29 MAs with 284 trials	ROR 0.9 (0.81,1.01)	
	pooled results of 7 empirical studies		ROR 0.82 (0.71,0.95)	Results very heterogeneous.
Wood 2008[16]	based on 3 other meta-epidemiological studies (Schulz, Kjaergard, Egger)	146 MAs with 1,346 trials	overall: 0.83 (0.74,0.93), n=102 reviews and 804 trials; subjective outcomes: 0.69 (0.59,0.82); objective outcomes: 0.91 (0.8,1.03)	Size of bias varied between MAs. Little difference for drug and non-drug interventions. No evidence of bias for all-cause mortality.
Unpublished Studies				
Als-Nielsen 2004[12]	included 6 empirical studies		ROR 0.79 (0.66,0.95)	Significant heterogeneity among studies.
Savovic 2008[17]	builds on Wood 2008 - same dataset with more studies	174 MAs	ROR 0.93 (0.89,0.96)	Less bias and little heterogeneity for mortality. Significant association and significant heterogeneity between MAs for other outcomes.
Nuesch 2008[18]	RCTs examining pain intensity in osteoarthritis	14 MAs with 163 trials and 40,436 patients	ES -0.16 (-0.33,0.02)	Estimates vary according to between-trial heterogeneity; effect more pronounced (and significant) in MAs with large between trial heterogeneity.

CI=confidence interval; MA=meta-analysis; ROR=ratio of odds ratio; AC=allocation concealment; DB=double-blinding; OR=odds ratio; RCT=randomized controlled trial; CL=Cochrane Library; ES=effect size

* Revised analyses published in 2008 showed significant association for AC (ROR 0.90 [0.82,0.995]).[50]

TABLE H2-C. EMPIRICAL EVIDENCE FOR BLINDING

Study	Sample	Size	Results (95% CI)	Comments
Published Studies				
Schulz 1995[8]	pregnancy and childbirth	33 MAs with 250 trials (62,091 participants; 12,030 outcome events)	ROR 0.83 (0.71,0.96) (adjusted for AC, SG, withdrawals)	Assessments based on reporting.
Moher 1998[9]	digestive, circulatory, mental health and pregnancy and childbirth	11 MAs with 127 trials (10,492 patients)	ROR 1.11 (0.76,1.63)	Majority of outcomes were objective.
Kjaergard 2001[10]	8 therapeutic areas: cardiology, surgery, pregnancy, schizophrenia, gynaecology, addictions, hypertension, neonatal	14 MAs with 190 RCTs (136,164 participants)	ROR 0.56 (0.33,0.98)*	Also compared large trials versus small trials with varying methodological quality, as well as small trials with varying methodological quality.
Balk 2002[13]	4 medical areas: cardiovascular, infectious disease, pediatrics, surgery	26 MAs with 276 trials	ROR 1.02 (0.79,1.24)	Quality is only one potential explanation for heterogeneity in treatment effect and should not be over-interpreted.
Egger 2003[14]	MAs in: infectious diseases, neonatology, neurology, obstetrics/gynaecology, psychiatry, other	45 MAs with 399 trials	ratio of pooled estimates 0.88 (0.75,1.04)	Some heterogeneity between MAs.

	pooled results from 4 empirical studies (Schulz, Moher, Kjaergard, Egger)		ratio of pooled estimates 0.86 (0.77,0.95)	
Siersma 2007[11]	MAs from Cochrane Library	523 trials from 41 reviews with 48 meta-analyses	ROR 0.92 (0.82,1.04)	Results consistent in terms of statistical significance for different statistical approaches.
Pildal 2007[15]	randomly selected MAs from PubMed	20 meta-analyses with 182 trials	ROR 0.94 (0.8,1.1)	Statistical interaction between DB and AC may exist.
	pooled results of 7 empirical studies		ROR 0.91 (0.83,1)	
Wood 2008[16]	based on 3 other meta-epidemiological studies (Schulz, Kjaergard, Egger)	146 MAs with 1346 trials	overall: 0.93 (0.83,1.04) n=76 MAs and 746 trials; objective outcomes: 1.01 (0.92,1.1); subjective outcomes: 0.75 (0.61,0.82); trials with AC: 1.02 (0.92,1.14), n=12 MAs with 60 trials	Overall heterogeneity significant. Inconsistencies in assessing DB across 3 included studies. Little difference for drug and non-drug interventions. No evidence of bias for all-cause mortality. No evidence of blinding as a source of bias for studies with adequate AC.
Unpublished Studies				
Als-Nielsen 2004[12]	included 6 empirical studies		ROR 0.82 (0.71,1.05)	Significant heterogeneity among studies.
Savovic 2008[17]	builds on Wood 2008 - same dataset with more studies	101 MAs	overall: ROR 0.97 (0.92,1.01)	Significant heterogeneity overall and for other outcomes. No heterogeneity for all-cause mortality.

CI=confidence interval; MA=meta-analysis; ROR=ratio of odds ratio; AC=allocation concealment; SG=sequence generation; DB=double-blinding; RCT=randomized controlled trial

* Revised analyses published in 2008 showed no significant association for DB (ROR 1.02 [0.94,1.11]).[50]

TABLE H2-D. EMPIRICAL EVIDENCE FOR MISSING DATA

Study	Sample	Size	Characteristic	Results (95% CI)	Comments
Published Studies					
Schulz 1995[8]	pregnancy and childbirth	33 MAs with 250 trials (62,091 participants; 12,030 outcome events)	reported exclusions	ROR 1.07 (0.94,1.21) (adjusted for AC, SG, DB)	
Kjaergard 2001[10]	8 therapeutic areas: cardiology, surgery, pregnancy, schizophrenia, gynaecology, addictions, hypertension, neonatal	14 MAs with 190 RCTs (136,164 participants)	follow-up (number and reasons for drop-outs and withdrawals described)	ROR 1.50 (0.80,2.78)	Also compared large trials versus small trials with varying methodological quality, as well as small trials with varying methodological quality.
Balk 2002[13]	4 medical areas: cardiovascular, infectious disease, pediatrics, surgery	26 MAs with 276 trials	drop-outs recorded	ROR 1.26 (0.87,2.05); n=141	Quality is only one potential explanation for heterogeneity in treatment effect and should not be over-interpreted.
			reasons for drop-outs given	ROR 0.93 (0.77,1.13); n=141	
			percentage of drop-outs	ROR 1.02 (0.94,1.12); n=261	
			ITT	ROR 0.91 (0.70,1.13); n=276	

Porta 2007[25]	two group RCTs identified in PubMed that performed both ITT and PP analyses on the primary endpoint	74 RCTs with binary outcomes	ITT versus per protocol	PP provides higher estimates of effect on average; unpredictability of bias in either direction; ITT more conservative but not necessarily better.	Analyses need to account for both random and non-random missingness. Neither ITT nor PP optimal by itself.
Siersma 2007[11]	MAs from Cochrane Library	523 trials from 41 reviews with 48 meta-analyses	ITT	ROR 1.01 (0.93,1.11)	Results consistent in terms of statistical significance for different statistical approaches.
Tierney 2005[23]	therapeutic questions in cancer	14 MAs of IPD with 133 trials and 21,905 patients	post-randomization exclusions versus ITT	No consistent effect at trial level (results changed in both directions); for MA, non-ITT analyses favoured treatment (p=0.03)	
Unpublished Studies					
Als-Nielsen 2004[12]	pooled results from 2 empirical studies		ITT	ROR 1.06 (0.92,1.22)	Significant heterogeneity among studies.
Nuesch 2008[27]	RCTs examining pain intensity in osteoarthritis	14 MAs with 172 trials and 39,298 patients	ITT	ES -0.09 (-0.23,0.05)	Estimates vary according to between-trial heterogeneity: significant when large between-trial heterogeneity.
Abraha 2008[26]	Trials published in 3 general and 3 specialised medical journals	223 trials	"modified" ITT versus ITT	ROR 0.85 (0.81,0.88)	

CI=confidence interval; MA=meta-analysis; ROR=ratio of odds ratio; AC=allocation concealment; SG=sequence generation; DB=double-blinding; RCT=randomized controlled trial; ITT=intention-to-treat; PP=per protocol; IPD=individual patient data; ES=effect size

TABLE H2-E. EMPIRICAL EVIDENCE FOR SELECTIVE OUTCOME REPORTING

Study	Sample	Size	Bias	Results (95% CI)	Comments
Published Studies					
Melander 2003[24]	trial reports submitted to the Swedish drug regulatory authority versus publications	42 placebo controlled studies of 5 selective serotonin reuptake inhibitors	ITT versus per protocol	majority presented only more favourable per protocol analysis	
Hahn 2002[33]	protocols submitted to local ethics committee	27 completed projects; 18 were published; 15 reports obtained (only 2 RCTs)	within-study selective reporting	RCTs each reported 5 outcomes not specified in protocol, most of which were statistically significant in favour of treatment over control.	Lack of detailed outcome definitions in protocols was problematic.
Williamson 2005[57]	SRs, from a previous project, where publication bias identified as potential problem	9 MAs with strong indication of publication bias	within-study selective reporting	case by case	Impact on conclusions of MA was minimal. In some cases, funnel plot asymmetry explained by selective outcome reporting.
Chan 2004[35]	randomized trials from ethics committees in Denmark in 1994-95; had to have at least one identifiable journal article; excluded abstracts	102 trials with 122 publications and 3,736 outcomes	completeness of reporting: statistically significant versus nonsignificant	efficacy OR 2.4 (1.4-4.0), n=50 trials; harm OR 4.7 (1.8-12.0), n=18 trials	No effect by funding source, sample size, number of study centers. Association between statistical significance and completeness of reporting

	and reports of preliminary findings		consistency of primary outcome btw protocol and publication	primary outcome changed, introduced or omitted in 62% of trials	varied widely between studies.
Chan 2004[34]	Protocols approved for funding by CIHR from 1990 to 1998	48 trials with 68 publications and 1,402 outcomes	completeness of reporting: statistically significant versus nonsignificant	efficacy OR 2.7 (1.5-5.0), n=30 trials; harm OR 7.7 (0.5-111), n=4 trials	
			consistency of primary outcome btw protocol and publication	primary outcome changed in 40% of trials	
Chan 2005[36]	trials published in Dec 2000 and identified through PubMed	519 trials with 553 publications and 10,557 outcomes	completeness of reporting	efficacy OR 2.0 (1.6-2.7), n=161 trials; harm OR 1.9 (1.1-3.5), n=43 trials	Exploratory analyses: multicentre trials associated with less bias; papers with definitions of primary outcomes associated with more bias
Furukawa 2007[38]	SRs with minimum 10 trials from CL 2005	156 SRs with 4,222 trials	selective outcome reporting	median 46% of trials contributed to MA; when outcomes favoured the intervention, effect estimate decreased with increasing proportion of trials in MA	

Dwan 2008[29]	SR of studies examining pub bias (n=11) and outcome reporting bias (n=5)		significant outcomes more likely to be fully reported	ORs ranged from 2.2 to 4.7 (n=3 studies)	Reasons for not reporting pre-specified outcomes included lack of clinical importance and lack of statistical significance.
			consistency of primary outcome	changed, introduced or omitted from protocol to publication in 40-62% studies	
Unpublished Studies					
Von Elm 2006[29;37]	drug trials submitted to university ethics committee in Switzerland from 1988 to 1998	451 trials	completeness of reporting	associated with statistical significance[29]	Minimal details and data reported in abstract.
Ghersi 2006[58]	ethics committee in Sydney from 1992 to 1996	103 published trials	consistency and completeness of reporting	consistency of primary outcomes from protocol to publication and completeness of reporting for primary and other outcomes was associated with completeness of sample size calculations; statistical significance associated with reporting all comparisons	

CI=confidence interval; ITT=intention-to-treat; RCT=randomized controlled trial; SR=systematic review; MA=meta-analysis; OR=odds ratio; CIHR=Canadian Institutes for Health Research; CL=Cochrane Library

TABLE H2-F. EMPIRICAL EVIDENCE FOR "OTHER SOURCES OF BIAS" RELEVANT TO PRESENT PROPOSAL

Study	Sample	Size	Bias	Results (95% CI)	Comments
Trials Stopped Early for Benefit					
Montori 2005[40]	SR of trials stopped early for benefit to Nov 2004	143 trials	stopped early for benefit	median RR 0.53 (0.28,0.66), n=126; trials with fewer events showed greater treatment effect: OR 28 (11-73)	typically industry funded (pharmacological interventions) in cardiology (acute coronary syndromes), cancer (lung cancer), and HIV/AIDS
Influence of Trial Sponsor					
Gotzsche 2006[45]	industry-initiated trials approved by ethics committee in Denmark	44 trials	sponsor has access to data	36% (16 trials)	Constraints by sponsors were rarely declared in publications.
			sponsor can stop trial	36% (16 trials)	
			constraints by sponsor on publication	91% (40 trials); in 50% sponsor owned data and/or had to approve manuscript	
Small Sample Size					
Juni 2008[47] (unpublished)	RCTs examining pain intensity in osteoarthritis	13 MAs with 156 trials and 37,594 patients	small (<200 patients) versus large trials	difference in ES: -0.23 (-0.37,-0.09)	Estimates vary according to between-trial heterogeneity: significant when large between-trial heterogeneity.

CI=confidence interval; SR=systematic review; RR=risk ratio; OR=odds ratio; RCT=randomized controlled trial; ES=effect size

APPENDIX H-3

CHILD FILTER

#1 (Infant* OR infancy OR Newborn* OR Baby* OR Babies OR Neonat* OR Preterm* OR Prematur* OR Postmatur* OR Child* OR Schoolchild* OR School age* OR Preschool* OR Kid or kids OR Toddler* OR Teen* OR Boy* OR Girl* OR Minors* OR Pubert* OR Pubescen* OR Prepubescen* OR Pediatric* OR Paediatric* OR Peadiatric* OR Nursery school* OR Kindergar* OR Primary school* OR Secondary school* OR Elementary school* OR High school* OR Highschool*):ti,ab,kw or (Adolesc*):ti,ab or (Infant OR Child OR Minors OR Puberty OR Pediatrics OR Schools):kw in Cochrane Reviews, Other Reviews and Clinical Trials

#2 adolescent*:kw in Cochrane Reviews, Other Reviews and Clinical Trials

#3 (adolescent* and (adult* or elderly or "middle aged" or "aged, 80 and over")):kw in Cochrane Reviews, Other Reviews and Clinical Trials

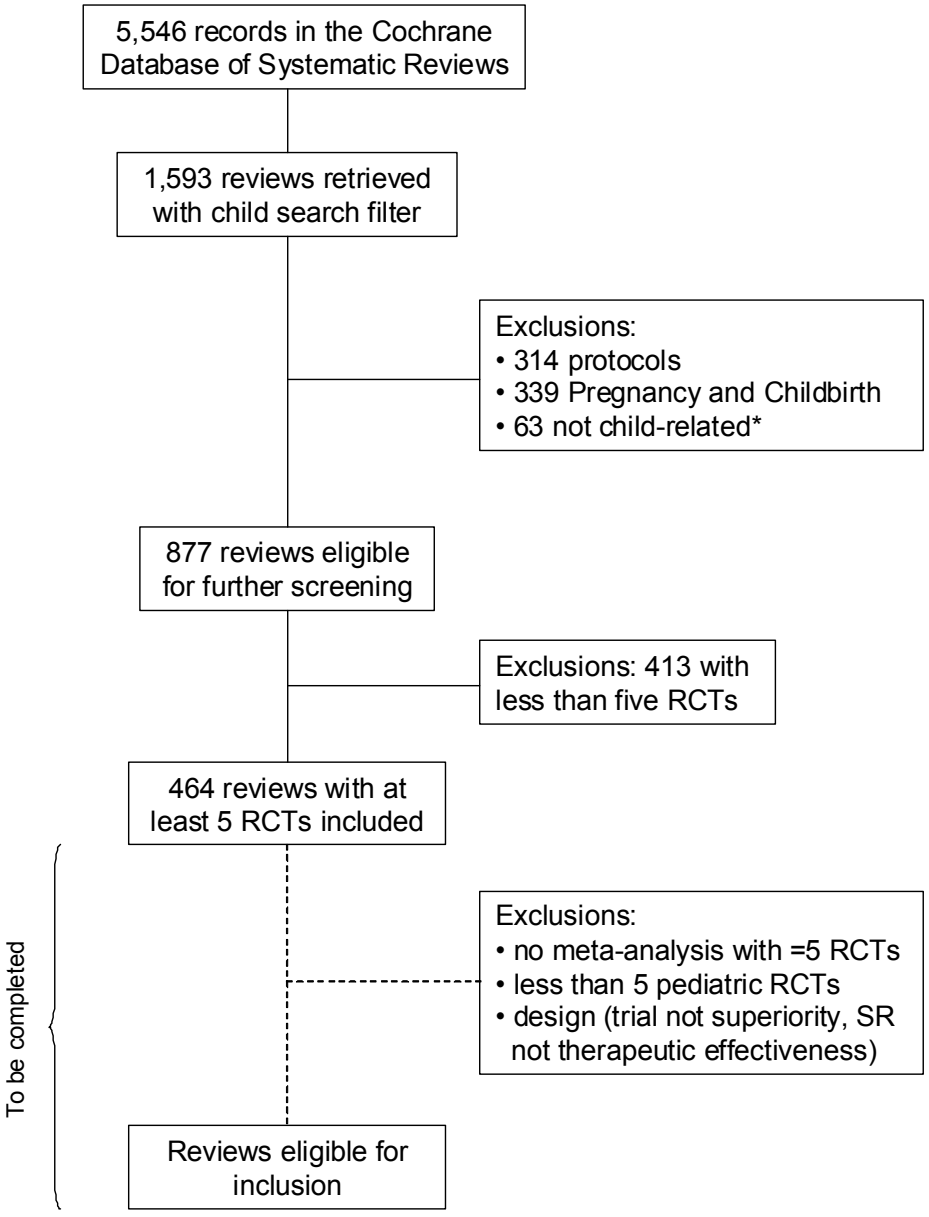
#4 (#3 AND NOT #1)

#5 (#1 OR #2)

#6 (#5 AND NOT #4)

APPENDIX H-4

FLOW DIAGRAM OF REVIEWS THROUGH SCREENING PROCESS



* (i.e., dementia, prostatic diseases, fertility regulation, title specifically stated adult only)

Potentially child-relevant systematic reviews identified from the Cochrane Database of Systematic Reviews, presented by Collaborative Review Group

Cochrane Collaborative Review Group	Number of Potentially Child-Relevant Reviews	Number with at least 5 trials
Neonatal	249	102
Airways	107	61
Acute Respiratory Infections	64	34
Developmental, Psychosocial and Learning Problems	49	29
Infectious Diseases	45	34
Cystic Fibrosis and Genetic Disorders	37	7
Oral Health	27	13
Injuries	25	14
Ear, Nose and Throat Disorders	24	14
Epilepsy	23	8
Renal	18	14
Skin	15	11
HIV/AIDS	14	9
Incontinence	12	9
Pain, Palliation and Supportive Care	12	8
Metabolic and Endocrine Disorders	11	6
Depression, Anxiety and Neurosis	11	7
Anaesthesia	10	6
Eyes and Vision	10	1
Neuromuscular	10	9
Tobacco Addiction	9	5
Inflammatory Bowel Disease and Functional Bowel Disorders	8	4
Heart	8	5
Musculoskeletal	7	5
Gynaecological Cancer	7	5
Wounds	7	6
Back	6	6
Bone, Joint and Muscle Trauma	6	4
Consumers and Communication	6	3
Effective Practice and Organisation of Care	6	4
Drugs and Alcohol	5	5
Peripheral Vascular Diseases	5	1
Colorectal Cancer	4	3
Movement Disorders	4	1
Schizophrenia	4	3
Upper Gastrointestinal and Pancreatic Diseases	4	2
Haematological Malignancies	3	2
Menstrual Disorders and Subfertility	3	2
Hepato-Biliary	2	2

APPENDIX H-5

CRITERIA FOR CATEGORIZATION OF VARIABLES

A. Classification of study reports as published or unpublished

Published: full or short reports, editorials, or letters appearing in a journal or journal supplement

Unpublished: all other reports

From: Egger M, Juni P, Bartlett C, Holenstein F, Sterne J. How important are comprehensive literature searches and the assessment of trial quality in systematic reviews? Empirical study. Health Technol Assess 2003; 7(1):1-76.

B. Classification of study outcomes as objective or subjective

"The definition of objective and subjective outcomes was based on the extent to which outcome assessment could be influenced by investigators' judgment. Objectively assessed outcomes included all cause mortality, measures based on a recognised laboratory procedure (such as measurement of haemoglobin concentrations), other objective measures (such as preterm birth), and surgical or instrumental outcomes (all of these were concerned with childbirth, such as caesarean section or instrumental delivery). Note that such surgical outcomes (classified as objectively assessed) depend on doctors' decisions, which could, in the absence of blinding, be affected by knowledge of the intervention received. Subjectively assessed outcome measures included patient reported outcomes, physician assessed disease outcomes (such as vascular events, pyelonephritis, or respiratory distress syndrome), measures combined from several outcomes, and withdrawals or study dropouts."

From: Wood L, Egger M, Gluud LL, Schulz KF, Juni P, Altman DG, Gluud C, Martin RM, Wood AJG, Sterne JAC. Empirical evidence of bias in treatment effect estimates in controlled trials with different interventions and outcomes: meta-epidemiological study. BMJ 2008;336(7644): 601-5.

C. Classification of funding source

Government, pharmaceutical industry, private, other, unclear.

From: Klassen TP, Wiebe N, Russell K, Stevens K, Hartling L, Craig WR, Moher D. Abstracts of randomized controlled trials presented at the society for pediatric research meeting: an example of publication bias. Arch Pediatr Adolesc Med 2002;156(5):474-9.

D. Classification of outcomes by completeness of reporting

Levels of Outcome Reporting		Reported Data	Data Sufficient for Inclusion in Meta-analysis
Full		No. of participants per group Effect size Precision or precise *P* value for continuous data*	Yes
Incomplete			
	Partial	Effect size or precision (± sample size and/or *P* value)†	No
	Qualitative	*P* value (± sample size)†	No
	Unreported	None	No

*Precise *P* value enables the calculation of the standard error if the treatment effect and sample sizes are given.
†Items in parentheses indicate "optional" data, i.e., those not necessary or not sufficient on their own to meet the requirements for the particular definition.

From: Chan AW, Hrobjartsson A, Haahr MT, Gotzsche PC, Altman DG. Empirical evidence for selective reporting of outcomes in randomized trials. Comparison of protocols to published articles. JAMA 2004;291:2457-2465.

E. Classification of interventions and controls

Intervention: drugs; rehabilitation or psychosocial; prevention or screening; surgery or radiotherapy; communication, organisational, or educational; alternative therapeutic; other.

From: Wood L, Egger M, Gluud LL, Schulz KF, Juni P, Altman DG, Gluud C, Martin RM, Wood AJG, Sterne JAC. Empirical evidence of bias in treatment effect estimates in controlled trials with different interventions and outcomes: meta-epidemiological study. BMJ 2008;336: 601-5.

Control: no intervention, placebo, or active intervention.

From: Siersma V, Als-Nielsen B, Chen W, Hilden J, Gluud LL, Gluud C. Multivariable modelling for meta-epidemiological assessment of the association between trial quality and treatment effects estimated in randomized clinical trials. Statistics in Medicine 2007;26:2745-58.

APPENDIX H-6

CRITERIA FOR JUDING RISK OF BIAS IN THE 'RISK OF BIAS' ASSESSMENT TOOL

From: Higgins JPT, Green S (editors). Cochrane Handbook for Systematic Reviews of Interventions *Version 5.0.1 (Table 8.4.a) [updated September 2008]. The Cochrane Collaboration, 2008. Available from www.cochrane-handbook.org.*

SEQUENCE GENERATION **Was the allocation sequence adequately generated? [Short form: *Adequate sequence generation*?]**	
Criteria for a judgement of 'YES' (i.e. low risk of bias).	The investigators describe a random component in the sequence generation process such as: • Referring to a random number table; • Using a computer random number generator; • Coin tossing; • Shuffling cards or envelopes; • Throwing dice; • Drawing of lots; • Minimization*. *Minimization may be implemented without a random element, and this is considered to be equivalent to being random.
Criteria for the judgement of 'NO' (i.e. high risk of bias).	The investigators describe a non-random component in the sequence generation process. Usually, the description would involve some systematic, non-random approach, for example: • Sequence generated by odd or even date of birth; • Sequence generated by some rule based on date (or day) of admission; • Sequence generated by some rule based on hospital or clinic record number. Other non-random approaches happen much less frequently than the systematic approaches mentioned above and tend to be obvious. They usually involve judgement or some method of non-random categorization of participants, for example: • Allocation by judgement of the clinician; • Allocation by preference of the participant; • Allocation based on the results of a laboratory test or a series

	of tests; • Allocation by availability of the intervention.
Criteria for the judgement of 'UNCLEAR' (uncertain risk of bias).	Insufficient information about the sequence generation process to permit judgement of 'Yes' or 'No'.

ALLOCATION CONCEALMENT **Was allocation adequately concealed? [Short form: *Allocation concealment*?]**	
Criteria for a judgement of 'YES' (i.e. low risk of bias).	Participants and investigators enrolling participants could not foresee assignment because one of the following, or an equivalent method, was used to conceal allocation: • Central allocation (including telephone, web-based, and pharmacy-controlled, randomization); • Sequentially numbered drug containers of identical appearance; • Sequentially numbered, opaque, sealed envelopes.
Criteria for the judgement of 'NO' (i.e. high risk of bias).	Participants or investigators enrolling participants could possibly foresee assignments and thus introduce selection bias, such as allocation based on: • Using an open random allocation schedule (e.g. a list of random numbers); • Assignment envelopes were used without appropriate safeguards (e.g. if envelopes were unsealed or nonopaque or not sequentially numbered); • Alternation or rotation; • Date of birth; • Case record number; • Any other explicitly unconcealed procedure.
Criteria for the judgement of 'UNCLEAR' (uncertain risk of bias).	Insufficient information to permit judgement of 'Yes' or 'No'. This is usually the case if the method of concealment is not described or not described in sufficient detail to allow a definite judgement – for example if the use of assignment envelopes is described, but it remains unclear whether envelopes were sequentially numbered, opaque and sealed.

BLINDING OF PARTICIPANTS, PERSONNEL AND OUTCOME ASSESSORS

Was knowledge of the allocated interventions adequately prevented during the study? [Short form: *Blinding*?]

Criteria for a judgement of 'YES' (i.e. low risk of bias).	Any one of the following: • No blinding, but the review authors judge that the outcome and the outcome measurement are not likely to be influenced by lack of blinding; • Blinding of participants and key study personnel ensured, and unlikely that the blinding could have been broken; • Either participants or some key study personnel were not blinded, but outcome assessment was blinded and the non-blinding of others unlikely to introduce bias.
Criteria for the judgement of 'NO' (i.e. high risk of bias).	Any one of the following: • No blinding or incomplete blinding, and the outcome or outcome measurement is likely to be influenced by lack of blinding; • Blinding of key study participants and personnel attempted, but likely that the blinding could have been broken; • Either participants or some key study personnel were not blinded, and the non-blinding of others likely to introduce bias.
Criteria for the judgement of 'UNCLEAR' (uncertain risk of bias).	Any one of the following: • Insufficient information to permit judgement of 'Yes' or 'No'; • The study did not address this outcome.

INCOMPLETE OUTCOME DATA

Were incomplete outcome data adequately addressed? [Short form: *Incomplete outcome data addressed*?]

Criteria for a judgement of 'YES' (i.e. low risk of bias).	Any one of the following: • No missing outcome data; • Reasons for missing outcome data unlikely to be related to true outcome (for survival data, censoring unlikely to be introducing bias); • Missing outcome data balanced in numbers across intervention groups, with similar reasons for missing data across groups; • For dichotomous outcome data, the proportion of missing outcomes compared with observed event risk not enough to have a clinically relevant impact on the intervention effect estimate; • For continuous outcome data, plausible effect size (difference in means or standardized difference in means) among missing outcomes not enough to have a clinically relevant impact on observed effect size; • Missing data have been imputed using appropriate methods.
Criteria for the judgement of 'NO' (i.e. high risk of bias).	Any one of the following: • Reason for missing outcome data likely to be related to true outcome, with either imbalance in numbers or reasons for missing data across intervention groups; • For dichotomous outcome data, the proportion of missing outcomes compared with observed event risk enough to induce clinically relevant bias in intervention effect estimate; • For continuous outcome data, plausible effect size (difference in means or standardized difference in means) among missing outcomes enough to induce clinically relevant bias in observed effect size; • 'As-treated' analysis done with substantial departure of the intervention received from that assigned at randomization; • Potentially inappropriate application of simple imputation.
Criteria for the judgement of 'UNCLEAR' (uncertain risk of bias).	Any one of the following: • Insufficient reporting of attrition/exclusions to permit judgement of 'Yes' or 'No' (e.g. number randomized not stated, no reasons for missing data provided); • The study did not address this outcome.

SELECTIVE OUTCOME REPORTING

Are reports of the study free of suggestion of selective outcome reporting? [Short form: *Free of selective reporting*?]

Criteria for a judgement of 'YES' (i.e. low risk of bias).	Any of the following: • The study protocol is available and all of the study's pre-specified (primary and secondary) outcomes that are of interest in the review have been reported in the pre-specified way; • The study protocol is not available but it is clear that the published reports include all expected outcomes, including those that were pre-specified (convincing text of this nature may be uncommon).
Criteria for the judgement of 'NO' (i.e. high risk of bias).	Any one of the following: • Not all of the study's pre-specified primary outcomes have been reported; • One or more primary outcomes is reported using measurements, analysis methods or subsets of the data (e.g. subscales) that were not pre-specified; • One or more reported primary outcomes were not pre-specified (unless clear justification for their reporting is provided, such as an unexpected adverse effect); • One or more outcomes of interest in the review are reported incompletely so that they cannot be entered in a meta-analysis; • The study report fails to include results for a key outcome that would be expected to have been reported for such a study.
Criteria for the judgement of 'UNCLEAR' (uncertain risk of bias).	Insufficient information to permit judgement of 'Yes' or 'No'. It is likely that the majority of studies will fall into this category.

OTHER POTENTIAL THREATS TO VALIDITY

Was the study apparently free of other problems that could put it at a risk of bias? [Short form: *Free of other bias*?]

Criteria for a judgement of 'YES' (i.e. low risk of bias).	The study appears to be free of other sources of bias.
Criteria for the judgement of 'NO' (i.e. high risk of bias).	There is at least one important risk of bias. For example, the study: • Had a potential source of bias related to the specific study design used; or • Stopped early due to some data-dependent process (including a formal-stopping rule); or • Had extreme baseline imbalance; or • Has been claimed to have been fraudulent; or • Had some other problem.
Criteria for the judgement of 'UNCLEAR' (uncertain risk of bias).	There may be a risk of bias, but there is either: • Insufficient information to assess whether an important risk of bias exists; or • Insufficient rationale or evidence that an identified problem will introduce bias.