Palliative and Supportive Care (2017), **15**, 20–31.

doi:10.1017/S1478951516000316

The needs of siblings of children with a life-threatening illness, part 2: Psychometric validation of the IBesFEMS

MARIANNE OLIVIER D'AVIGNON, PH.D.,[1] SERGE DUMONT, PH.D.,[2] PIERRE VALOIS, PH.D.,[1] AND S. ROBIN COHEN, PH.D.[3,4]

[1]Faculté des Sciences de l'Éducation, Université Laval, Québec, Canada
[2]École de Service Social, Université Laval, Québec, Canada
[3]Departments of Oncology and Medicine, McGill University, Montreal, Québec, Canada
[4]Lady Davis Institute, Québec, Canada

(RECEIVED July 20, 2015; ACCEPTED March 29, 2016)

ABSTRACT

Objective: Life-threatening illnesses in children have a significant impact on the lives of their brothers and sisters. Consequently, special attention must be paid to the specific needs of these siblings to help them cope with their situations. To address this issue, we developed an inventory of the needs of the adolescent siblings of severely ill children, the Inventaire des Besoins de la Fratrie d'Enfants Malades Sévèrement (IBesFEMS) [Needs Inventory for Siblings of Critically Ill Children]. The present article describes a preliminary validation study of this new instrument.

Method: In a prospective cohort study, the 48-item instrument was administered via a website or paper to 58 siblings.

Results: Our study revealed that the measure has an estimated internal consistency of 0.96 and a temporal stability intraclass correlation coefficient (*ICC*) of 0.86 ($p < 0.01$). Its convergence validity is also satisfactory.

Significance of results: Our findings suggest that the IBesFEMS is highly relevant for pediatric palliative care clinicians and researchers. Future studies should investigate its factorial structure and predictive validities.

KEYWORDS: Needs, Siblings, Pediatric, Palliative care, Questionnaire development

INTRODUCTION

It is difficult to estimate the number of children with a life-threatening illness or condition due to the variable trajectories of such illnesses and the uncertainty of their prognoses (Hynson & Sawyer, 2001; Lenton et al., 2006). Lenton and his collaborators (2006) put the prevalence of noncancerous life-threatening illness among children between the ages of 0 and 19 at 1.5 per 1,000. This suggests that nearly 11,800 Canadian children are afflicted with a disease with a low survival rate (Statistics Canada, 2014). That number is just over 13,200 when the incidence of pediatric cancer is taken into account (Canadian Cancer Society, 2014).

Life-threatening illnesses and the demands they create have considerable repercussions for the siblings of afflicted children, notably by disrupting their daily lives and monopolizing the attention and availability of their parents (Jones et al., 2011; Gaab et al., 2013). In light of this, special attention must be paid to the specific needs of siblings to help them cope with their situations.

This is the second of two articles on the findings of a research initiative to develop and validate an inventory of the needs of siblings of children with a life-threatening illness (the IBesFEMS; see the Supplementary Material). While the first article

Address correspondence and reprint requests to: Marianne Olivier d'Avignon, Faculté des sciences de l'éducation, Pavillon des sciences de l'éducation, Université Laval, Québec, Canada G1 V 0A6. E-mail: Marianne.davignon@fse.ulaval.ca.

described the various steps involved in the conceptualization and subsequent content validation of the IBesFEMS (Olivier d'Avignon et al., 2016), this article discusses its initial psychometric validation.

Siblings in Pediatric Palliative Care

Life-threatening illnesses among children, as well as their consequences, create considerable disruptions in family life and in the roles played by family members. Pediatric palliative care has developed gradually since the 1980s as a means to alleviate the suffering of these young patients (Armstrong-Dailey & Zarbock, 2009). The goal of this care is not only to accompany the child whose life is threatened, but also to ease the multidimensional suffering of the entire family, be it physical, social, spiritual, or psychological in nature (van Horne & Kautz, 2007; McNamara-Goodger & Feudtner, 2012). The family-centered approach is an important characteristic of this care philosophy (Jones et al., 2011; Contro & Scofield, 2012). The care team's interventions respect the primordial role of the parents and siblings in accompanying the seriously ill child, and the team tends to play a supportive role (Lenton et al., 2006; Price et al., 2013). Although increasing attention has been paid to sibling well-being and support in clinical intervention plans, the lack of evidence in this emerging field of research suggests the need for a serious research effort to support practice development (Institute of Medicine [IOM], 2003; Sourkes et al., 2005; Ministère de la Santé et des service sociaux du Québec [MSSS], 2006; Stevenson et al., 2013).

Numerous private and public organizations have called for more scientific research to document the specific needs of siblings (Whittam, 1993; Robinson & Mahon, 1997; IOM, 2003; Wilson et al., 2003; Contro & Scofield, 2006; Graham et al., 2006; Jones, 2006; MSSS, 2006) so as to better orient medical and psychosocial practices (IOM, 2003). The lack of measurement instruments specifically aimed at siblings and tailored to the various stages of their development has been deplored (IOM, 2003; Graham et al., 2006; MSSS, 2006). A recent questionnaire developed by an Australian team, the Sibling Needs Cancer Instrument (SNCI) (Patterson et al., 2011; 2014), is an interesting initiative. However, it addresses only the siblings of children with cancer, whereas palliative care practitioners care for children with a broad spectrum of life-threatening illnesses (Feudtner et al., 2011). The IBesFEMS was designed for siblings of children with various life-threatening illnesses and has the added advantage of assessing siblings' perceived levels of satisfaction with the degree to which their needs are met as well as the importance of each need.

Despite the emergence of a care philosophy for pediatric palliative care, there has been very little research documenting the experience of siblings, and knowledge about their specific needs is fragmented (Jones, 2006; MSSS, 2006; Contro & Scofield, 2006; 2012; Long et al., 2015). Professionals involved in this practice context therefore have very few theoretical referents and limited means with which to assess the needs of these children in the family or care environment. The typology of needs used to develop the IBesFEMS is a theoretical contribution that allows practitioners to identify the specific needs of this population (Olivier d'Avignon et al., 2016). The findings from part 1 of this study demonstrate that the content validity of the IBesFEMS is very good. The present study (part 2) tests its reliability and convergent and criterion validities, along with its sensitivity to social desirability bias, in adolescent siblings of children with various life-threatening illnesses.

Conceptual Framework

The IBesFEMS is conceptually based on a typology of the needs of the siblings of children with a life-threatening illness. This typology played an important role in the development of the new instrument by paving the way for a detailed description of the study concept. Readers are invited to consult the first article in this series (Olivier d'Avignon et al., 2016) for the conceptual and operational definitions associated with the concepts used in this study and for a description of the different steps involved in the development of the IBesFEMS. The various psychometric validation procedures are based both on classical test theory and on the conception of validity outlined in the updated edition of *The Standards for Educational and Psychological Testing*, a document drafted jointly by the American Educational Research Association [AERA], the American Psychological Association [APA], and the National Council on Measurement in Education [NCME] (2014). The present paper describes the evidence collected to establish its psychometric properties, for both clinical identification of important unmet needs and research. Validity refers here to the degree to which this evidence, once juxtaposed with the theoretical elements of the field under study, supports the interpretation of the results stemming from the uses proposed for the IBesFEMS (Streiner & Norman, 2008).

METHOD

Sample

The target population consisted of teenagers aged 12 to 18 years with a brother or sister older than 1 year

https://doi.org/10.1017/S1478951516000316 Published online by Cambridge University Press

Table 1. *IBesFEMS*[a] *item and response scale*

	Is this need important to you?				Do you feel this need is being met?			
	Not important (1)	Somewhat important (2)	Important (3)	Very important (4)	Not at all (1)	Somewhat (2)	Yes (3)	Very much (4)
I need my parents to keep me informed about my brother's (or sister's) illness.	1	2	3	**4**	**1**	2	3	4

[a] Inventaire des Besoins de la Fratrie d'Enfants Malades Sévèrement (IBesFEMS) [Needs Inventory for Siblings of Critically Ill Children].

(including a stepbrother or stepsister, to whom they may or may not be related by blood) diagnosed with a life-threatening illness. In addition, the teenagers must have been able to speak, read, and write in French and reside in Canada. Adolescents suffering from a life-threatening illness, mental retardation, or a psychiatric disorder were excluded from our study. Some 61 participants were recruited between September of 2009 and May of 2010 using a nonprobabilistic sampling procedure, with the collaboration of four healthcare establishments in the province of Québec, Canada, offering pediatric palliative care.[1] Prior approval was obtained from each of the collaborating establishments' research ethics committees. Parents signed a consent form, while teenagers were required to sign an assent form.

Needs Construct

The main variable of interest in our study is need. Viewed here from a clinical perspective, it is defined as the difference between an actual and a desired state (Baldwin, 1998). The IBesFEMS is a questionnaire made up of 48 items grouped into 10 measurement dimensions (Olivier d'Avignon et al., 2016). Some of the measurement dimensions of the instrument include only one item. During the content validation phase, other items had been proposed, but, despite different wording, these items were deemed redundant (Olivier d'Avignon et al., 2016). The importance attributed to each of these needs as well as the degree of need satisfaction was rated by respondents using a 4-point Likert-type scale. Using an algorithm allows us to obtain, for each item, a single score, by combining the two subscales (Olivier d'Avignon et al., 2016). This single score is referred to here as the Unmet Need Score (UNS). The algorithm employed is $UNS = I * (4 - S)$. The higher the UNS, the more the item with which it is associated indicates an important unmet need, that is, important, but not satisfied. Table 1 illustrates one IBesFEMS item and its response scale. In this example, a UNS of 12 is obtained by combining a score of 4 on the importance subscale and a score of 1 on the satisfaction subscale ($12 = 4 * (4-1)$), witch indicates an important unmet need. Item UNS values can range from 0 (all needs completely satisfied) to 12 (all needs important and unsatisfied). The total score on the IBesFEMS is obtained by summing all UNSs and can theoretically range from 0 to 576. A high score indicates a large number of unmet important needs. Table 1 illustrates one IBesFEMS item and its response scale.

Validity

Convergent Validity

Convergent validity was tested by generating evidence based on the relationship between the needs construct as measured by the IBesFEMS and three other constructs with which there is a theoretically recognized link (Streiner & Norman, 2008; AERA et al., 2014).

It has been found that siblings who receive support from peers, friends, family, and teachers cope better with a life-threatening illness and that their needs are better met (Woodgate, 1999; Sloper, 2000; Read et al., 2011; Lapwood & Goldman, 2012). The first variable was social support, measured using the abridged French version of the Social Support Questionnaire (SSQ–6) (Sarason et al., 1983). This questionnaire has acceptable psychometric properties in both its original English version (Sarason et al., 1983) and the French version (Rascle et al., 1997). A high score indicates an elevated level of availability of and satisfaction with social support (Rascle et al., 1997). For this psychometric study,

[1]The healthcare establishments that collaborated on this study were the Centre Hospitalier Universitaire (CHU) Sainte-Justine, the Centre Mère-Enfant du Centre Hospitalier Universitaire de Québec (CHUQ), the Centre Hospitalier Universitaire de Sherbrooke (CHUS), and Le Phare: Enfants et Familles, a pediatric hospice.

https://doi.org/10.1017/S1478951516000316 Published online by Cambridge University Press

we anticipated moderate negative correlations ($-0.49 < r < -0.30$) between results obtained with the IBesFEMS and those generated using the SSQ–6 (de Vaus, 2002*b*), so that siblings with more social support would report fewer unmet important needs.

Several studies have reported that siblings of children with a life-threatening illness are at risk of developing psychological problems (Sharpe & Rossiter, 2002; Laufersweiler-Plass et al., 2003; Barlow & Ellard, 2006; Long et al., 2013). This risk is greater when parents are physically and emotionally unavailable to attend fully to the needs of the healthy siblings (Alderfer et al., 2010). Psychological distress was assessed using the Indice de Détresse Psychologique de Santé-Québec [Psychological Distress Index, used in the Québec Health Survey] (IDPSQ–14), an abridged French version of the Psychological Symptom Index (PSI) (Ilfeld, 1976). This instrument has acceptable psychometric properties (Deschesnes, 1998). The total score for the IDPSQ–14 ranges from 14 to 56. The higher the score, the more severe the sibling's symptoms of psychological distress (Deschesnes, 1998). For our psychometric study, we anticipated a strong positive correlation ($0.50 < r < 0.69$) between the results obtained with the IBesFEMS and those generated using the IDPSQ–14 (de Vaus, 2002*b*).

Siblings of children with cancer can feel jealousy and anger toward a seriously ill child (Nolbris et al., 2006; Woodgate, 2006). Elevated negative emotions could be related to unmet needs with respect to information and a normal social life for the siblings (Houtzager et al., 2005). The guilt and shame these thoughts trigger can adversely affect the self-esteem of these young people (Packman et al., 2005). Therefore, the self-esteem of participants was assessed using the French translation of Rosenberg's Self-Esteem Scale (RSES) (Vallières & Vallerand, 1990). This instrument has been validated with a sample of college students and has acceptable psychometric properties (Vallières & Vallerand, 1990). Scores can vary between 10 and 40. The higher the result, the higher the respondent's self-esteem. For our psychometric study, we anticipated moderate negative correlations ($-0.49 < r < -0.30$) between IBesFEMS and RSES scores (de Vaus, 2002*b*). Evaluation of the convergent validity of the IBesFEMS with respect to the SSQ–6, IDPSQ–14, and RSES was assessed using Spearman's rank correlation coefficient (r_s).

Criterion Validity

Criterion validity or known-group comparison is a second type of validation that provides a means to generate evidence based on external variables (AERA et al., 2014). This is done by dividing the sample into subgroups based on a characteristic recognized in the literature as contributing to differentiated experiences on the variable the new instrument purports to measure, and then comparing scores. In our study, siblings' perceptions of whether or not their daily lives were disrupted by the illness were selected to create two subgroups. In order to measure this variable, the respondents were asked, "Over the past two weeks, do you think that your sibling's illness changed your daily life?" and the two subgroups were differentiated by their response (yes/no). The Mann–Whitney (U) test was utilized to assess whether the groups had significantly different IBesFEMS scores.

Reliability

Internal Consistency and Item Analysis

The internal consistency of an instrument reflects the overall homogeneity of the instrument and each of its measurement dimensions (DeVellis, 2012) and is calculated using Cronbach's alpha (α). Analysis of the homogeneity index of each versus all items as well as their discriminatory power provided additional information on this aspect of reliability. The analysis indicates the coherence between an item and the whole, as well as any redundancy between items, the latter of which could artificially inflate the value of α. A delicate balance between homogeneity and specificity is sought in item analysis (Sabourin et al., 2005). This information was obtained using item–total and interitem correlation.

Temporal Stability (Test–Retest Reliability)

Finally, to perform a test–retest analysis, a sibling subsample completed the IBesFEMS a second time two weeks later. A multiple-choice transitional question was posed to ensure that the sick child's condition was still the same as at the time of the initial testing: "Since the last time you completed this questionnaire, has your brother (or sister's) health (1) deteriorated, (2) improved, or (3) remained the same?" A copy of the instrument was mailed to the first 29 brothers and sisters who agreed to take part in this second testing. Seven questionnaires were disregarded, as there had been a change in the sick child's condition since the first testing. The total number of retests used in the analysis was therefore 22. The temporal stability of the total results with the IBesFEMS from these two tests was analyzed using the intraclass correlation coefficient (*ICC*; Deyo and colleagues [1991]).

Social Desirability Bias

Sensitivity to the phenomenon of social desirability was verified using Spearman's rank order correlation

https://doi.org/10.1017/S1478951516000316 Published online by Cambridge University Press

(r_s) between the abridged French version of the Balanced Inventory of Desirable Responding (BIDR–6 abridged) (Paulhus, 1991) and the IBesFEMS. This questionnaire has two subscales: the self-deception subscale offers an internal consistency index of 0.75, while the other-deception subscale has an index of 0.70 (Frenette et al., 2000).

Data Analysis Procedure

Three participants responded to less than 53% of IBesFEMS items. Since this meant there was a significant amount of missing data, the questionnaires from these subjects were rejected. Further, only one measurement item among all the completed measurement instruments had more than 5% missing data. In order to reduce the negative impact of the missing data, the expectation–maximization (EM) data imputation method was selected (de Vaus, 2002*a*; Tabachnick & Fidell, 2006) and applied to the five measurement instruments used.

As suggested by de Vaus (2002*a*) and Munro (2005), the normality of the distribution was analyzed using the Kolmogorov–Smirnov test for all data, in each instrument, with the exception of the results obtained in the retest, which were analyzed using the Shapiro–Wilk test, since the number of participants was less than 50. Statistical analyses for the study were conducted using the Statistical Package for Social Sciences (SPSS, v. 13.0).

Analysis of the normality of the distribution of these tests revealed an abnormal distribution for the data collected from all instruments, with the exception of the following questionnaires: the SSQ–6 (availability subscale), the RSES, and the BIDR–6. Consequently, all of the statistical analyses conducted were nonparametric. A probability of type I error with $p < 0.05$ was considered statistically significant.

RESULTS

Participants and Mode of Completion

The total number of participants in this study was 58. A total of 22 (36.1%) participants completed the questionnaire online on a secure website. Since some were reluctant to use this means of data collection, a paper version of the questionnaire was mailed to those who expressed a preference. Some 39 (63.9%) participants chose the paper version. The teenagers were asked to complete the questionnaires on their own, without any help. They reported that it took them between 30 and 60 minutes to perform this task. Figure 1 shows the number of participants recruited in relation to the number of families approached, refusals, and withdrawals. It also illustrates the sample distribution, based on source of referrals, among the four different pediatric care facilities that helped recruit participants.

The average age was 14.3 ($SD = 1.76$), with a range of 12 to 18. The number of males and females was comparable (52.6% female and 47.4% male). The mean number of months since the sick child's diagnosis was 72.91 ($SD = 74.05$), and the observed median number of months since the sick child's diagnosis was 73. The nature and frequency of diagnoses are shown in Table 2. Some 51 teenagers (89.5%) affirmed that their ill sibling required special care or attention at home, while 39 (67.2%) perceived the illness as having had an impact on their daily lives within the previous two weeks. Lastly, 12 (20.7%) participants who completed the questionnaire expressed a desire to meet with a professional for help in coping with the serious illness afflicting their brother or sister. They were subsequently referred to specialized healthcare professionals.

Convergent Validity

The correlation between the IBesFEMS and psychological distress ($r_s = 0.577$, $p < 0.01$) was as expected. The correlation between the IBesFEMS and perceived social support ($r_s = -0.273$, $p < 0.05$) was slightly lower than anticipated. A moderate association was found between the availability and satisfaction dimensions of social support. As reported in the literature, this association seems to be particularly significant between satisfaction with received social support and unmet needs, in this case as measured by the IBesFEMS (Rascle et al., 1997). As predicted, a strong association was found between psychological distress symptoms and IBesFEMS scores. However, convergence with the RSES did not yield the expected results. The association between self-esteem and IBesFEMS scores proved to be low and insignificant (Table 3).

Criterion Validity

A Mann–Whitney U test was employed to compare two respondent subgroups divided on the basis of whether the illness caused a disruption in their daily lives. There was no significant difference between the means of the two groups: disruption ($n = 39$, $M = 114.90$, $SD = 89.60$); no disruption ($n = 19$, $M = 97.26$, $SD = 75.77$; $U = 0.782$, $p > 0.05$).

Internal Consistency

Internal consistency was calculated using Cronbach's alpha for the entire instrument and for each of the measurement dimensions with more than one item. All the values of Cronbach's α obtained

https://doi.org/10.1017/S1478951516000316 Published online by Cambridge University Press

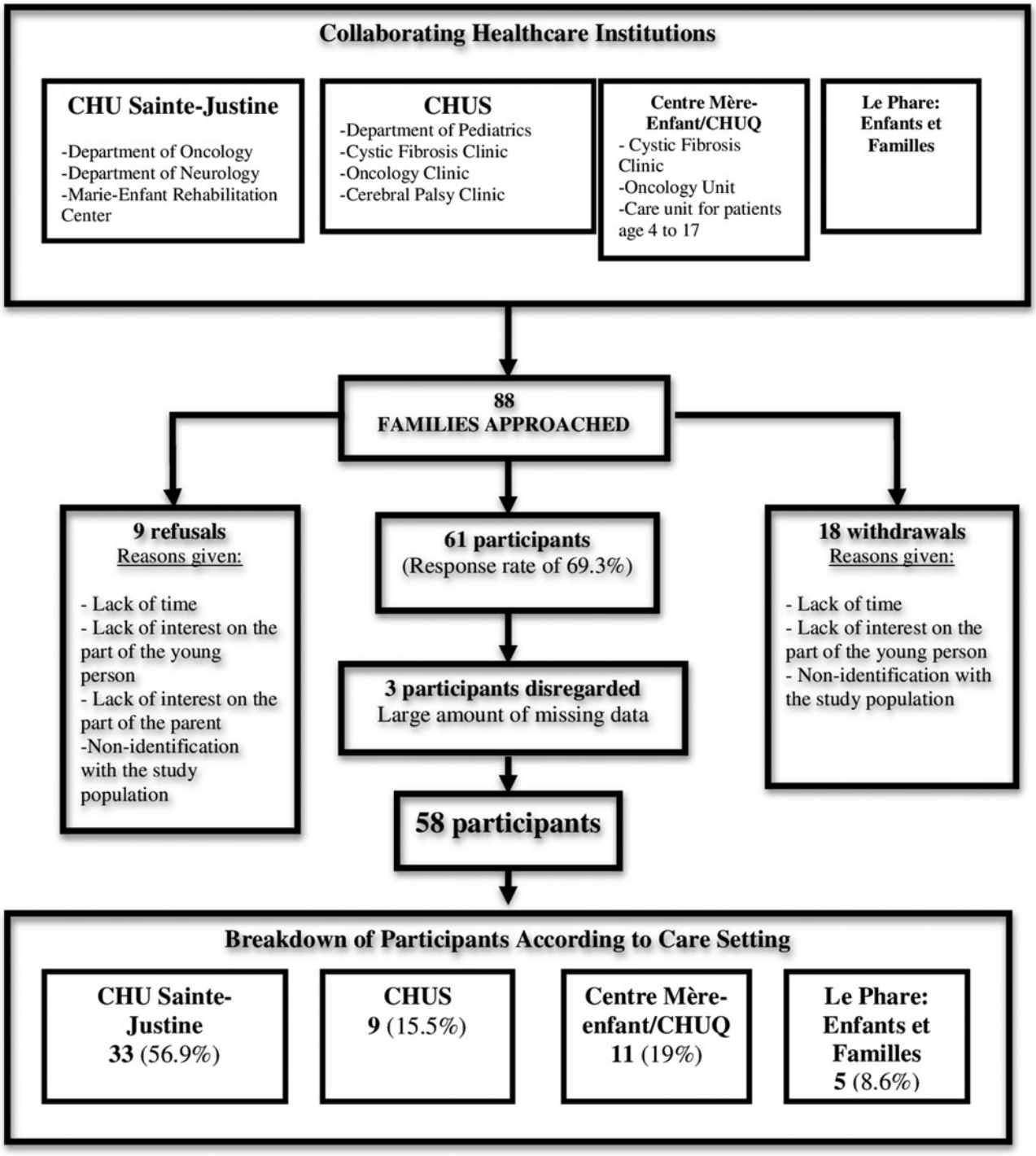

Fig. 1. Data collection flowchart.

met the desired threshold for a new measure ($\geq$0.70) (Nunnally & Bernstein, 1994), with the exception of the dimension "relationship with significant adults," which has two items ($r = 0.60$) (Table 4).

All but one of the corrected item–total correlations obtained were above the recommended threshold of 0.30 (de Vaus, 2002*a*). Moreover, for each of the questionnaire items, when ignored, Cronbach's α ranged from 0.959 to 0.961 for item analysis by dimension as well as for the entire instrument. This would suggest that all the items contributed equally to measurement of the construct (Streiner & Norman, 2008). Only item 4, "I need someone from the hospital to come to my classroom to explain my brother (or sister's) illness," did not reach the desired threshold, with an item–total correlation of $r_{it} = 0.22$.

https://doi.org/10.1017/S1478951516000316 Published online by Cambridge University Press

Table 2. *Nature and frequency of the diagnoses*

Sick child's diagnosis	Frequency (%)
Cancer	27 (46.6%)
Severe encephalopathy and other diseases of the central nervous system	12 (20.7%)
Rare syndromes and congenital defects	9 (15.5%)
Cystic fibrosis	6 (10.3%)
Muscular dystrophy	2 (3.4%)
Chronic kidney disease	1 (1.7%)
Cardiopathy	1 (1.7%)

Analysis of the interitem correlations can help to detect item redundancy. Streiner and Norman (2008) recommend a moderate interitem correlation no greater than 0.70, so as to confirm each item's real discriminatory power. The average interitem correlation for the IBesFEMS is 0.34, and the average interitem correlation for each dimension ranges from 0.30 to 0.46. However, four item pairs show interitem correlations slightly above the desired threshold: items 32 and 21 (0.73), 26 and 41 (0.71), 2 and 5 (0.72), and 31 and 32 (0.79). These items were retained because in the previous content validity study a consultation with experts (siblings, psychometricians, palliative care researchers, pediatric palliative care clinicians) did not find those items to be redundant (Olivier d'Avignon et al., 2016) and because they are not extremely high (>0.90).

Temporal Stability

The *ICC* for the IBesFMES completed 2 weeks apart, when the ill child's condition was perceived as stable, was 0.86 ($p < 0.01$).

Social Desirability Bias

The sensitivity of the IBesFEMS to social desirability bias was assessed using a Spearman's correlation test between the data obtained with the IBesFEMS and that obtained with the abridged version of the BIDR–6. These findings showed a negative correlation ($r_s = -0.322$, $p < 0.05$). This suggests that the participants who expressed a high level of unmet needs on the IBesFEMS are not at all or only slightly likely to be influenced by such a bias, and, inversely, participants expressing a low level of unmet needs are more likely to do so to please the evaluator, suggesting potential underreporting of unmet need.

DISCUSSION

In the pediatric palliative research and care community, there is a need for an instrument to measure the needs of siblings of children with a life-threatening illness (IOM, 2003; MSSS, 2006). The IBesFEMS was designed to identify and assess the needs of the brothers and sisters of children with a life-threatening illness. The procedure followed for psychometric validation of the initial version of the instrument complied with the recommended standards in psychometry and allowed us to affirm that the IBesFEMS satisfies the validity and reliability criteria recognized by the AERA, APA, and NCME.

The results obtained in our study with the IBesFEMS were related to important variables. Analyses identified a strong and significant relationship between the presence of unmet needs among respondents and the presence of psychological distress. Analyses showed a moderate negative relationship with satisfaction with perceived social support, and a weak negative relationship with the availability of social support. These results confirm those reported with the SCNI (Patterson et al., 2011; 2014), which also detected a relationship between the presence of psychological distress among siblings and the presence of unmet needs. Our study also found the predicted relationship between both dissatisfaction with and nonavailability of social support and incidence of unmet needs. These results confirmed two of our hypotheses for convergent validity. In contrast, the correlation between the IBesFEMS and self-esteem was not significant, which was not predicted.

Table 3. *Spearman correlation coefficients between the IBesFEMS*[a] *and other questionnaires*

	SSQ–6[b] availability subscale	SSQ–6[b] satisfaction subscale	RSES[c]	IDPSQ–14[d]
IBesFEMS[a]	$r_s = -0.273$*	$r_s = -0.418$**	$r_s = 0.141$	$r_s = 0.577$**

[a] Inventaire des Besoins de la Fratrie d'Enfants Malades Sévèrement (IBesFEMS) [Needs Inventory for Siblings of Critically Ill Children].
[b] Social Support Questionnaire.
[c] Rosenberg's Self-Esteem Scale.
[d] Indice de Détresse Psychologique de Santé-Québec [Psychological Distress Index, used in the Québec Health Survey].
* $p < 0.05$. ** $p < 0.01$.

https://doi.org/10.1017/S1478951516000316 Published online by Cambridge University Press

Table 4. *Cronbach's α internal consistency index for the entire instrument and by dimension*

Components	Number of items	Unmet needs	Importance subscale	Satisfaction subscale
IBesFEMS[a]	48	0.96	0.93	0.96
Measurement dimensions				
School life	5	0.77	0.72	0.81
Relationship with peers	1	b	b	b
Recreation	1	b	b	b
Hospital setting	4	0.77	0.73	0.77
Relationship with the community	1	b	b	b
Relationship with significant adults	2	0.60	0.76	0.74
Family life	7	0.76	0.72	0.79
Relationship with parents	7	0.80	0.74	0.83
Ontosystem	12	0.89	0.82	0.89
Relationship with sick child	8	0.88	0.80	0.83

[a] Inventaire des Besoins de la Fratrie d'Enfants Malades Sévèrement (IBesFEMS) [Needs Inventory for Siblings of Critically Ill Children].
[b] Not applicable because the dimension has only one item.

The known-group comparison between the two subgroups differentiated by impact of the illness on their daily lives was not significant. This may be because the IBesFEMS is not sufficiently sensitive to pick up a difference in needs. An alternative explanation is the high number of children who contracted a chronic disease since birth ($n = 26$, 45%) among the sick children. Siblings of these sick children have been exposed to their brother or sister's illness for many years, and in some cases since birth. Their daily lives may have been constructed around the presence of the life-threatening illness. Despite this, some may have responded in the affirmative about the impact of the illness on daily life. It is possible, however, that in this group of respondents, the teens, or their parents, have implemented coping mechanisms to allow for optimal functioning by healthy siblings, thereby diminishing their unmet needs. In contrast, children with cancer are likely to have had the illness for a shorter period of time. Consequently, siblings and family have less time to adjust. In this regard, an exploratory analysis of the results from the IBesFEMS appears to show a difference between the siblings of children with cancer and siblings of children with other types of life-threatening illnesses. Cancer group siblings reported more unmet needs ($n = 27$, $U = 288.5$, $p < 0.05$), and they also perceived perturbations in their daily lives in greater proportion ($\chi^2 = 14.7$, $p < 0.01$) than the noncancer group. Sample heterogeneity is one factor that could explain some of these results. Nonetheless, the idea of introducing a generic instrument that reflects the diverse clientele in pediatric palliative care has the added advantage of being consistent with the norms of clinical practice in the field. With a view to further verifying the instrument's sensitivity to discriminate between respondents, it would be interesting in the course of subsequent studies to determine whether the IBesFEMS detects a difference according to where the sick child is situated on the illness trajectory (e.g., diagnosis, relapse, end of life). According to the literature, illness trajectory has an impact on the emotional state and needs of siblings (Freeman et al., 2003).

All but one of the findings related to reliability meet the recommended thresholds. The internal consistency index associated with the dimension "relationship with significant adults," which has only two items, is slightly lower. It is worth noting that a lesser number of items in a dimension decreases internal consistency reliability (DeVellis, 2012). This measurement dimension is important for the instrument because it allowed us to measure needs expressed by sibling participants during the IBesFEMS development phase (Olivier d'Avignon et al., 2016), and it was therefore retained. The inter-item correlations for these two items show some results above 0.40, but lower than the threshold of 0.70. This indicates convergence of both items 13 and 14 with certain other items, but without establishing redundancy, which allows us to postulate that this dimension measures different needs and must remain distinct. This hypothesis will have to be confirmed by a factor analysis to verify whether these two items are naturally associated with other dimensions. Such an analysis was not performed as part of the current study due to the limited number of participants in the sample. To perform factor analysis, we recommend an absolute sample size of at least 100 subjects and a 10:1 ratio (McCallum et al., 1999).

The reliability characteristics of the instrument obtained by analyzing item homogeneity and discriminatory power showed that slight changes would

https://doi.org/10.1017/S1478951516000316 Published online by Cambridge University Press

need to be made to the current version of the IBesFEMS. For instance, to make item 4 more homogenous with the other items in the "school" dimension, the item "I need someone from the hospital healthcare team to come to my class to explain the illness of my brother or my sister" could be replaced with "I need the nurse at my school to come to my class to explain the illness of my brother or my sister." In addition, the discriminatory power of several items could be increased, notably of items 31 and 32, which deal, respectively, with the need for reassurance about the risk of loved ones becoming sick and the siblings themselves becoming sick. These items show an interitem correlation of 0.79, which is above the 0.70 threshold (Streiner & Norman, 2008). Given that these two items measure convergent needs, it is possible that the proximity of the items in the questionnaire contributed to the higher correlation. One possible solution could be to separate the two items. It would be worthwhile to repeat the analyses of the reliability characteristics discussed here in a larger sample. In addition to enabling factor analysis and construct validity, this would make results more robust and reduce the risk of measurement error (DeVellis, 2012). Validation of an English version of the IBesFEMS is also a forthcoming step that will allow for wider dissemination and use of this measurement instrument. Lastly, sensitivity to change is a characteristic that helps verify a measurement instrument's capacity to accurately detect meaningful (rather than statistically significant) changes (Beaton et al., 2001). It will be important to verify this characteristic because of the IBesFEMS's projected use in different clinical situations. It is possible that the instrument will be used on more than one occasion with the same sibling in order to track the evolution of their needs based on their development, the trajectory of their illness, or the assistance they have received.

In light of the largely positive results obtained in our psychometric validation, it is appropriate to recommend that the IBesFEMS be employed in clinical and research settings with teenage siblings of children with a life-threatening illness. Special attention will, however, have to be paid to items 4, 31, and 32, which will be slightly modified.

In the present study, analysis of the relationship between the results obtained with the IBesFEMS and the SSQ–6 suggest that unmet needs are more likely to be related to dissatisfaction with perceived social support rather than to the absence of individuals available to help. This finding points to the importance of educating and raising awareness among the loved ones of siblings as to their needs. Given that parents tend to be the primary providers of support for a sick child's healthy siblings, various authors have stressed the importance of helping to prepare parents to meet the needs of these children as effectively as possible (Hashemi & Shokrpour, 2010). As such, the IBesFEMS could not only help enhance parents' awareness of the general needs of the child's siblings, but it could also be used to identify individual needs. This suggests an area for future research.

Certain characteristics distinguish the IBesFEMS questionnaire. First, the items have a strong theoretical basis because of the typology of needs from which they were developed (Olivier d'Avignon et al., 2016). The contribution of Bronfenbrenner's human bioecological theory made possible the categorization of needs in 10 different living environments. This classification considers not only all spheres of sibling life—it also identifies who they would like to have fill the need. The authors of the SCNI, the only similar instrument, interviewed siblings as well as employees of an Australian support organization about their perceived needs. In results published in 2011, 10 domains of needs were identified for this population. These domains, generated by a thematic analysis, seem to be general categories of needs and do not seem to be based on theoretical or conceptual foundations. This remains the case for the seven domains of the final version of SCNI proposed in 2014, which are statistically derived (Patterson et al., 2014). In addition, temporal stability is a characteristic evaluated in the context of validation of the present study. The authors of the SCNI report a test–retest correlation coefficient similar to an *ICC* of 0.88. However, the authors do not mention if they controlled for the stability of the ill child's health condition. Finally, and perhaps most important, the IBesFEMS not only measures unmet needs—it also evaluates how important each is and thus allows all actors involved in caring for siblings to focus on their most important unmet needs.

CONTRIBUTION TO CLINICAL PRACTICE AND RESEARCH

The availability of the IBesFEMS can help initiate changes in clinical practice by providing a better understanding of the needs of siblings. It will allow healthy brothers and sisters to express themselves on the importance of their needs and the perceived satisfaction of those needs. This standardized measurement instrument would provide a means to personalize psychosocial interventions by taking into account wide-ranging individual variability with regard to needs and enabling interventions that are better targeted to a sibling's specific situation (Jones et al., 2011). Moreover, the instrument would make it possible to conduct ongoing assessment of the needs of an individual over time. The utilization of this

https://doi.org/10.1017/S1478951516000316 Published online by Cambridge University Press

questionnaire might have an indirect effect in potentially improving the effectiveness of interventions in this population by targeting individualized unmet needs. Through the same indirect pathway, intervention at the level of unmet needs could alleviate unnecessary suffering among ill siblings. More effective interventions are likely to have a beneficial effect on seriously ill children or teenagers who are worried about the well-being of other family members (Whittam, 1993; IOM, 2003; Wilson et al., 2003). This new tool could help fill a need for clinical assessments. Another step would be to explore how it could be integrated into practice and whether this would have a positive effect on certain outcomes.

The IBesFEMS has the advantage of being a generic questionnaire that can be employed in families who have a child with a life-threatening illness, regardless of the diagnosis. In a context in which clinicians working in pediatric palliative care are faced with a broad range of life-threatening illnesses and conditions, the versatile nature of this new instrument certainly has its merits. However, before making the IBesFEMS available in care settings, a minimal clinically important difference (MCID) must be estimated. The MCID could be defined as the smallest difference in score on response to their needs that siblings perceive as beneficial (Copay et al., 2007). Determining this threshold would enable healthcare professionals to better interpret IBesFEMS final scores and assess the urgency of taking action and seeking professional help for siblings. The IBesFEMS's internal homogeneity appears to show that it is precise enough to be utilized in individual assessment. DeVellis (2012) notes that a minimum internal consistency α of 0.90 is recommended for overall internal consistency of measurement instruments destined for clinical use.

A second contribution that the IBesFEMS could make is related to the development of research in the field. It is hoped that the instrument would help generate knowledge about the needs of a little-studied population. Research findings could provide evidence that could be employed to develop innovative clinical practices. There appears to be an important need for such evidence, as the research and clinical pediatric palliative care are emerging domains and practice standards are still in the early stages (Graham et al., 2006; Wolfe & Siden, 2012). This kind of evidence would help to promote a family-centered approach, a recommendation supported by numerous authors (Lewis & Prescott, 2006; Jones et al., 2011; Lapwood & Goldman, 2012).

Finally, this measurement instrument will make it possible to assess existing programs for the siblings of children with a life-threatening illness. The impact of these programs could be documented and improvements made to them so that they better meet siblings' specific needs.

CONCLUSION

Siblings of children with a life-threatening illness have important needs that deserve special attention, not only from their parents and communities, but also from clinicians and researchers in the field. With the IBesFEMS it is now possible to quantitatively assess the needs of this population. One of the original contributions from our study is the theoretical role of the typology of needs. Comprised of needs in 10 living environments, this typology was developed qualitatively with siblings and parents. The IBesFEMS is a measurement instrument that is ready for use. At the same time, it can be employed to collect data to further evaluate its psychometric properties and better understand the meaning of changes in score. The results obtained during its development and validation phases are certainly promising.

ACKNOWLEDGMENTS

Dr. Olivier d'Avignon is grateful for the support of a doctoral fellowship provided by a joint initiative of the Canadian Institutes of Health Research and the Canadian Cancer Society Research Institutes: the Strategic Training Program in Palliative Care Research. The study was also funded by other doctoral fellowships provided by (1) the Social Sciences and Humanities Research Council of Canada and (2) Fonds de Recherche Société et Culture de Québec.

SUPPLEMENTARY MATERIALS AND METHODS

To view supplementary material for this article please visit http://dx.doi.org/10.1017/S1478951516000316.

REFERENCES

Alderfer, M.A., Long, K.A., Lown, E.A., et al. (2010). Psychosocial adjustment of siblings of children with cancer: A systematic review. *Psycho-Oncology, 19*(8), 789–805.

American Educational Research Association (AERA), American Psychological Association (APA) & National Council on Measurement in Education (NCME) (2014). *Standards for educational and psychological testing*. Washington, DC: American Educational Research Association.

Armstrong-Dailey, A. & Zarbock, S. (2009). *Hospice care for children*. 3rd ed. New York: Oxford University Press.

Baldwin, S. (1998). *Needs assessment and community care: Clinical practice and policy making*. Oxford: Butterworth–Heinemann.

Barlow, J.H. & Ellard, D.R. (2006). The psychosocial well-being of children with chronic disease, their parents and siblings: An overview of the research evidence

base. *Child: Care, Health and Development*, *32*(1), 19–31.
Beaton, D.E., Bombardier, C., Katz, J.N., et al. (2001). A taxonomy for responsiveness. *Journal of Clinical Epidemiology*; *54*(12), 1204–1217.
Canadian Cancer Society (2014). *Canadian cancer statistics, 2014*. Toronto: Canadian Cancer Society.
Contro, N. & Scofield, S. (2006). The power of their voices: Child and family assessment in pediatric palliative care. In *Oxford textbook of palliative care for children*. A. Goldman et al. (eds.), pp. 143–153. New York: Oxford University Press.
Contro, N. & Scofield, S. (2012). The power of their voices: Assessing the child and the family. In *Oxford textbook of palliative care for children*, 2nd ed. A. Goldman et al. (eds.), pp. 58–67. New York: Oxford University Press.
Copay, A.G., Subach, B.R., Glassman, S.D., et al. (2007). Understanding the minimum clinically important difference: A review of concept and methods. *The Spine Journal*, *7*, 541–546.
Deschesnes, M. (1998). Étude de la validité et de la fidélité de l'indice de détresse psychologique de santé Québec (IDPSQ–14), chez une population adolescente [Assessment of the validity and reliability of Santé Québec's psychological distress index (IDPSQ–14) in an adolescent population]. *Psychologie Canadienne*, *39*(4), 288–298.
de Vaus, D. (2002*a*). *Analyzing social science data: 50 key problems in data analysis*. Thousand Oaks, CA: Sage.
de Vaus, D. (2002*b*). *Survey in social research*, 5th ed. London: Routledge.
DeVellis, R.F. (2012). *Scale development: Theory and applications*, 3rd ed. Thousand Oaks, CA: Sage.
Deyo, R.A., Diehr, P. & Patrick, D. (1991). Reproducibility and responsiveness of health status measure statistics and strategies for evaluation. *Controlled Clinical Trials*, *12*, 142S–158S.
Feudtner, C., Hexem, K. & Rourke, M. (2011). Epidemiology and the care of children with complex conditions. In *Textbook of interdisciplinary pediatric palliative care*. J. Wolfe et al. (eds.), pp. 7–17. Philadelphia: Elsevier Saunders.
Freeman, K., O'Dell, C. & Meola, C. (2003). Childhood brain tumors: Children's and siblings' concerns regarding the diagnosis and phase of illness. *Journal of Pediatric Oncology Nursing*, *20*(3), 133–140.
Frenette, E., Valois, P., Lussier, Y., et al. (2000). *Validité de la version abrégée du questionnaire de désirabilité sociale de Paulhus* [Validity of the shortened version of Paulhus social desirability questionnaire]. Communication donnée au 68^{e} congrès de l'Association francophone pour le savoir (ACFAS). Montreal, Québec.
Gaab, E.M., Owens, G. & MacLeod, R.D. (2013). The voices of young New Zealanders involved in pediatric palliative care. *Journal of Palliative Care*, *29*(3), 186–192.
Graham, R., Dussel, V. & Wolfe, J. (2006). Research. In *Oxford textbook of palliative care for children*. A. Goldman et al. (eds.), pp. 615–647. New York: Oxford University Press.
Hashemi, F. & Shokrpour, N. (2010). The impact of education regarding the needs of pediatric leukemia patients' siblings on the parents' knowledge and practice. *The Health Care Manager*, *29*(1), 75–79.
Houtzager, B.A., Grootenhuis, M.A., Hoekstra-Weebers, J.E.H.M., et al. (2005). One month after diagnosis: Quality of life, coping and previous functioning in siblings of children with cancer. *Child: Care, Health and Development*, *31*(1), 75–87.
Hynson, J.L. & Sawyer, S.M. (2001). Paediatric palliative care: Distinctive needs and emerging issues. *Journal of Paediatrics and Child Health*, *37*(4), 323–325.
Ilfeld, F.W. (1976). Further validation of a psychiatric symptom index in a normal population. *Psychological Reports*, *39*, 1215–1228.
Institute of Medicine (IOM), Committee on Palliative and End-of-Life Care for Children and Families (2003). *When children die: Improving palliative and end of life care for children and families*. Washington, DC: The National Academic Press.
Jones, B.L. (2006). Companionship, control and compassion: A social work perspective on the needs of children with cancer and their families at the end of life. *Journal of Palliative Medicine*, *9*(3), 774–788.
Jones, B.L., Gilmer, M.J., Parker-Raley, J., et al. (2011). Parents' and siblings' relationships and the family experience. In *Textbook of interdisciplinary pediatric palliative care*. J. Wolfe et al. (eds.), pp. 135–147. Philadelphia: Elsevier Saunders.
Lapwood, S. & Goldman, A. (2012). Impact on the family. In *Oxford textbook of palliative care for children*, 2nd ed. A. Goldman et al. (eds.), pp. 117–129. New York: Oxford University Press.
Laufersweiler-Plass, C., Rudnik-Schöneborn, S., Zerres, K., et al. (2003). Behavioural problems in children and adolescents with spinal muscular atrophy and their siblings. *Developmental Medicine & Child Neurology*, *45*, 44–49.
Lenton, S., Goldman, A., Eaton, N., et al. (2006). Development and epidemiology. In *Oxford textbook of palliative care for children*. A. Goldman et al. (eds.), pp. 3–13. New York: Oxford University Press.
Lewis, M. & Prescott, H. (2006). Impact of life-limiting illness on the family. In *Oxford textbook of palliative care for children*. A. Goldman et al. (eds.), pp. 154–178. New York: Oxford University Press.
Long, K.A., Alderfer, M.A., Ewing, L.J., et al. (2013). The role of contextual threat in predicting self-reported distress among siblings of children with cancer. *Journal of Clinical Psychology in Medical Settings*, *20*(2), 199–208.
Long, K.A., Goldish, M., Lown, E.A., et al. (2015). Major lessons learned from a nationally based community–academic partnership to childhood cancer. *Families, Systems, & Health*, *33*(1), 61–67.
McCallum, R.C., Widamen, K.F., Zhang, S., et al. (1999). Sample size in factor analysis. *Psychological Methods*, *4*(1), 84–99.
McNamara-Goodger, K. & Feudtner, C. (2012). History and epidemiology. In *Oxford textbook of palliative care for children*, 2nd ed. A. Goldman et al. (eds.), pp. 3–11. New York: Oxford University Press.
Ministère de la Santé et des Services Sociaux du Québec [MSSS] (2006). *Normes en matière de soins palliatifs pédiatriques* [Québec standards of practice for pediatric palliative care]. Québec: Auteur.
Munro, B.H. (2005). *Statistical methods for health care research*, 5th ed. Philadelphia: Lippincott Williams & Wilkins.
Nolbris, M., Enskär, K. & Hellström, A.L. (2006). Experience of siblings of children treated for cancer. *European Journal of Oncology Nursing*, *11*, 106–112.
Nunnally, J.C. & Bernstein, I.H. (1994). *Psychometric theory*, 3rd ed. New York: McGraw-Hill.

Olivier d'Avignon, M., Dumont, S., Valois, P. et al. (2016). The needs of siblings of children with a life-threatening illness, part 1: Conceptualization and development of a measure. *Palliative & Supportive Care*. Manuscript submitted for publication, under review.

Packman, W., Greenhalgh, J., Chesterman, B., et al. (2005). Siblings of pediatric cancer patients: The quantitative and qualitative nature of quality of life. *Journal of Psychosocial Oncology*, *23*(1), 87–108.

Patterson, P., Millar, B. & Visser, A. (2011). The development of an instrument to assess the unmet needs of young people who have a sibling with cancer: Piloting the Sibling Cancer Needs Instrument (SCNI). *Journal of Pediatric Oncology Nursing*, *28*(1), 16–26.

Patterson, P., McDonald, F.E.J., Butow, P., et al. (2014). Psychometric evaluation of the Sibling Cancer Needs Instrument (SCNI): An instrument to assess the psychosocial unmet needs of young people who are siblings of cancer patients. *Supportive Care in Cancer*, *22*(3), 653–665.

Paulhus, D. (1991). Balanced inventory of desirable responding. In *Measure of personality and social psychological attitude*. J.P. Robinson et al. (eds.), pp. 17–60. New York: Academic Press.

Price, J., Jordan, J. & Prior, L. (2013). A consensus for change: Parent and professional perspectives on care for children at the end of life. *Issues in Comprehensive Pediatric Nursing*, *36*(1–2), 70–87.

Rascle, N., Aguerre, C., Bruchon-Schweitzer, M., et al. (1997). Soutien social et santé: Adaptation française du questionnaire de soutien social de Sarason, le S.S.Q. [Social support and health: Sarason's Social Support Questionnaire. French adaptation and validation]. *Les Cahiers Internationaux de Psychologie Sociale*, *33*, 35–51.

Read, J., Kinali, M., Muntoni, F., et al. (2011). Siblings of young people with Duchenne muscular dystrophy: A qualitative study of impact and coping. *European Journal of Paediatric Neurology*, *15*(1), 21–28.

Robinson, L. & Mahon, M. (1997). Sibling bereavement: A concept analysis. *Death Studies*, *21*, 477–499.

Sabourin, S., Valois, P. & Lussier, Y. (2005). L'utilisation de questionnaires en recherche: Une solution pratique qui nécessite une démarche rigoureuse [The use of questionnaires in research: A practical solution that requires a rigorous method]. In *Recherche psychosociale*, 2nd ed. S. Bouchard & C. Cyr (eds.), pp. 279–320. Québec: Presses de l'Université Québec.

Sarason, I.G., Levine, H.M., Basham, R.B., (1983). Assessing social support: The social support questionnaire. *Journal of Personality and Social Psychology*, *44*(1), 127–139.

Sharpe, D. & Rossiter, L. (2002). Siblings of children with a chronic illness: A meta-analysis. *Journal of Pediatric Psychology*, *27*(8), 699–710.

Sloper, P. (2000). Experiences and support needs of siblings of children with cancer. *Health & Social Care in the Community*, *8*(5), 298–306.

Sourkes, B., Frankel, L., Brown, M., et al. (2005). Food, toys and love: Pediatric palliative care. *Current Problems in Pediatric and Adolescent Health Care*, *35*(9), 350–386.

Statistics Canada (2014). *Population by sex and age group*. CANSIM, table 051–0001. Available from http://www.statcan.gc.ca/tables-tableaux/sum-som/l01/cst01/demo10a-eng.htm.

Stevenson, M., Achille, M. & Lugasi, T. (2013). Pediatric palliative care in Canada and the United States: A qualitative metasummary of the needs of patients and families. *Journal of Palliative Medicine*, *16*(5), 566–577.

Streiner, D.L. & Norman, G.R. (2008). *Health measurement scales: A practical guide to their development*, 4th ed. New York: Oxford University Press.

Tabachnick, B. & Fidell, L.S. (2006). *Using multivariate statistics*. Boston: Pearson/Allyn & Bacon.

Vallières, E.F. & Vallerand, R.J. (1990). Traduction et validation Canadienne-Française de l'échelle de l'estime de soi de Rosenberg [French Canadian translation and validation of the Rosenberg Self-Esteem Scale]. *International Journal of Psychology*, *25*, 305–316.

van Horne, E. & Kautz, D. (2007). Promotion of family integrity in the acute care setting. A review of the literature. *Dimensions of Critical Care Nursing*, *26*(3), 101–107.

Whittam, E. (1993). Terminal care of the dying child: Psychosocial implications of care. *Cancer*, *71*(Suppl. 10), 3450–3462.

Wilson, D.M., MacAdam, M., Olson, K., et al. (2003). Pediatric end-of-life care. *Final report. Integrated end-of-life care: A Health Canada synthesis research project*. Unpublished research report. Edmonton: Authors.

Wolfe, J. & Siden, H. (2012). Research in pediatric paediatric palliative care. In *Oxford textbook of palliative care for children*, 2nd ed. A. Goldman et al. (eds.), pp. 441–457. New York: Oxford University Press.

Woodgate, R.L. (1999). Social support in children with cancer: A review of the literature. *Journal of Pediatric Oncology Nursing*, *16*, 210–213.

Woodgate, R.L. (2006). Siblings' experiences with childhood cancer. *Cancer Nursing*, *29*(5), 406–414.