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Development of an international research agenda for adult congenital heart disease nursing

Eva Goossens MSc ^a, Desiree Fleck PhD ^{b,c}, Mary M. Canobbio MN ^d, Jeanine L. Harrison MSc ^e, Philip Moons PhD ^{a,f,g*} on behalf of the International Society of Adult Congenital Heart Disease (ISACHD) Nursing Network

^a Center for Health Services and Nursing Research, Katholieke Universiteit Leuven, Leuven, Belgium

^b Cardiac Center, Children's Hospital of Philadelphia, Pennsylvania, USA

^c University of Pennsylvania, School of Nursing, Philadelphia, Pennsylvania, USA

^d University of California, Los Angeles, School of Nursing, Ahmanson/UCLA Adult Congenital Heart Disease Center, Los Angeles, California, USA

^e Toronto Congenital Cardiac Centre for Adults, University Health Network, Toronto General Hospital, Division of Cardiology, Toronto, Canada

^f Division of Congenital and Structural Cardiology, University Hospitals Leuven, Leuven, Belgium

^g The Heart Centre, Copenhagen University Hospital, Copenhagen, Denmark

***Address for correspondence:**

Philip Moons, Center for Health Services and Nursing Research, Katholieke Universiteit Leuven, Kapucijnenvoer 35 box 7001, B-3000 Leuven, Belgium; Tel: +32-16-336984; Fax: +32-16-336970; E-mail: philip.moons@med.kuleuven.be

Abstract

Background

Since the population of adults with congenital heart disease (CHD) is growing, the role of nurse specialists is expanding. In order to advance ACHD nursing, the establishment of an international nursing research agenda is recommended. We aimed to investigate research priorities as perceived by nurse specialists and researchers in ACHD.

Methods

We applied a sequential quan-qual design. In the quantitative phase, a two-round Delphi study was conducted, in which 37 nurse specialists and nurse researchers in ACHD care participated. Respondents assessed the level of priority of 21 research topics using a 9-point rating scale (1=no priority at all; 9=very high priority). In the qualitative phase, semi-structured interviews were performed with six selected Delphi panelists, to scrutinize pending research questions.

Results

This study revealed that priority should be given to studies investigating knowledge and education of patients, outcomes of Advanced Practice Nursing, quality of life, transfer and transition, and illness experiences and psychosocial issues in adults with CHD. A low priority was given to post-operative pain, sexual functioning, transplantation in ACHD, and health care costs and utilization. Agreement about the level of priority was obtained for 14 out of 21 research topics.

Conclusion

Based on this study, we could develop an international research agenda for ACHD. Researchers ought to focus on these areas of highest priority, in order to expand and strengthen the body of knowledge in ACHD nursing.

Keywords: Heart defects, congenital; Nursing research; Delphi technique; Adult; Research priorities; Mixed Methods research design

1. Introduction

The number of adults with congenital heart disease (CHD) in society is continuously increasing (1). Based on a population study in Québec, Canada, it is estimated that the prevalence of CHD is 4.09 per 1000 adults (2). Extrapolating these figures to the approximated population of adult individuals (≥ 18 years) (esa.un.org/unpd/upp2008/all-wpp-indicators_components.htm), there are about 1 million and 2.4 million adult patients with CHD in the USA and in Europe, respectively.

Irrespective of the treatment received in early childhood, patients with CHD remain susceptible to develop medical complications, such as arrhythmias, ventricular dysfunction, endocarditis, and secondary pulmonary hypertension (1,3). They are also prone to experience psychosocial and behavioral problems (4,5) such as anxiety (5), depression (5), compromising health behavior (6) and declined social integration due to problems with employability and insurability (7-10). Specific issues such as reproduction and pregnancy, requires an increasing attention (11,12). Hence, life-long specialized care is recommended for most patients, to prevent and treat possible medical and non-medical problems (13,14). Furthermore, a significant proportion of patients is at risk for re-operation and thus need to be seen regularly in ACHD regional centers and followed for life (15,16). As a result, a series of task forces, consensus statements, and guidelines have been convened to better define the appropriate delivery of care, the practice and providers of care for this growing population of adult patients (15-21). Nursing has played a pivotal role in patient care, teaching and research since adult congenital heart disease (ACHD) programs were first described (22).

According to consensus documents produced by American, Canadian and European societies, nurses who are trained and educated in adult congenital heart disease (ACHD) care, play an

integral role of the interdisciplinary ACHD teams (18,20,23-27). Nurse specialists play a key role in assessing patients' needs, counseling patients and their families, and facilitating the transition and transfer of adolescents with CHD to adult care (4,13,22,27,28). Despite the acknowledged role ACHD nurses have made, the nursing aspects of care provided by ACHD nurse specialists is, however, not based on scientific evidence. Therefore, the need to develop a nursing research agenda in ACHD is pivotal to expand the current body of knowledge and furthermore to provide evidence-based ACHD nursing care (4,29). The primary focus of research conducted by nurses includes issues such as quality of life (30-38), illness experiences (39-47), knowledge and education (48-54), and transition and transfer to adult services (55-58).

To further advance the practice of ACHD nursing based on scientific merit, the development of an international nursing research agenda has been recommended (59). The purpose, of this study was to identify research priorities as perceived by nurse specialists and researchers in ACHD nursing.

2. Materials and methods

2.1 Study design

We used a sequential quan-qual design. This is a form of mixed methods research design, comprised of a quantitative (quan) phase, followed by a qualitative (qual) phase (60). In the quantitative phase, a Delphi study was conducted. A Delphi study is a structured consensus method that uses a group facilitation technique, consisting of multiple survey rounds. This technique is used when the goal is to transform the opinions of individual experts into a single group consensus (61-63). A panel of informed individuals, knowledgeable and experienced with the subject under investigation, is identified and compiled (61). These experts are sent a series of multiple structured questionnaires. Hence, the process of a Delphi study is multistaged. Results

from each round are reported descriptively to the participants in subsequent rounds. Presentation of the collective opinion provides the participant with additional information, through which the panelist can confirm or adjust the scores given in the preceding survey rounds. Data collection is completed when a predetermined level of consensus is reached within the participating panel (61). In the present study, we used the Delphi method to quantitatively assess the level of priority that ACHD nurse experts give to a set of predefined research topics. For the present study, the Delphi study consisted of two rounds in which electronic questionnaires were individually filled out by the participants.

Upon completion of the Delphi study, we initiated the qualitative phase. Semi-structured telephone interviews were conducted with panelists who participated in both preceding rounds of the Delphi study.

2.2 Sample

Potential participants for the Delphi survey were sought from the membership list of the International Society for Adult Congenital Heart Disease (ISACHD) Nursing Network (64). In addition, names of nursing participants at the 2007 ACHD congress in Philadelphia and personal contacts known to the authors were added. Finally, the clinic directory of the Adult Congenital Heart Association (www.achaheart.org) was screened to identify hospitals that had nurse specialists in their ACHD program. An email was sent to this list of individuals to identify potential participants. Eligibility criteria were (i) nurses whose clinical practice primarily focused on ACHD care; (ii) held a position as a nurse specialist, nurse practitioner or nurse researcher in ACHD; (iii) expressed their willingness to participate in this Delphi study; and (iv) availability of valid contact information. A total of 47 nurses met the inclusion criteria.

During the first Delphi round, 37 of the 47 eligible participants completed the survey form (response rate=78.7%). In the second round, 33 of the 37 nurses who participated in the first round, completed the form (response rate=89.2%). The characteristics of the participants (n=37) are described in **Table 1**.

Potential participants for the qualitative phase were selected based on the priority scores they individually gave to predetermined research topics during the Delphi study. Individual ratings of the level of priority were compared to the overall group rating. Potential interviewees were selected if their individual ratings were either divergent or convergent with the overall group rating. Furthermore, potential interviewees were included when they were actively involved in nursing research. A total of six out of eight potential interviewees (75%) agreed to participate.

2.3 Delphi survey instrument

The Delphi survey form was developed in two stages. First, a literature review was performed to determine the topics already investigated in nurse-led studies. We conducted a systematic literature search in the databases Medline and Cinahl, using the keywords ‘congenital heart’ AND adult AND nurs*. This search resulted in 330 hits (**Figure 1**), published between January 1980 and July 2010.

After elimination of duplicates, publications by non-nurses, and articles for which no abstract or full text was available, 159 publications were identified for further analysis. Overall, 101 articles pertained to ACHD, nine of which were editorials, eight were conference abstracts, and 84 were full articles. Forty-six articles were either reviews or reports, leaving 64 empirical studies for systematic analysis. The majority of these empirical nursing studies investigated quality of life (n=10) (30-38,62), followed by illness experiences and psychosocial issues (n=9)

(39-47), knowledge and education (n=7) (48-54), transfer and transition (n=4) (55-58), and health behavior (n=4) (66-69) (**Figure 1**). Issues that were investigated to a lesser extent were: pregnancy and gynaecological issues (11,12,70); organization of care (71,72); sexual functioning (73); implementation of advanced practice nursing (29); post-operative pain (74); nursing care in general (75); health care costs and utilization (76); physical activity (77); and palliative care (78). Based on the results of this systematic literature review, a preliminary list of 13 research topics in ACHD nursing was composed.

This preliminary list was then presented to the executive board of the ISACHD Nursing Network. The members of this board were asked to propose additional topics for nursing research. Eight additional topics were proposed, resulting in a total of 21 ACHD-related research topics. These 21 topics were included in the final survey form for round one of the Delphi study. Respondents were asked to indicate the level of priority for each of the 21 predefined research topics, using a 9 point-rating scale (1=no priority at all; 9=very high priority). Free text space was provided, giving participants the opportunity to generate ideas and suggest additional research topics based on their professional experiences. The survey form was accompanied by a short self-report questionnaire on professional characteristics.

2.4 Data collection

Eligible experts received an electronic invitation explaining the aims of the study and describing the methodological principles of a Delphi survey. A questionnaire was attached to this letter with instructions to be completed within two weeks. Non-responders received up to three reminders.

After data collection was completed in round 1, data were analyzed descriptively (median, Q1-Q3 and range).The panelists were then invited to partake in round two. In this

second round, all panelists were given their personal responses from round one, the overall group rating of all 21 predefined research topics. Furthermore, panelists were given the opportunity to reconsider their initial scores in light of the results of the preceding round. Again, in the second Delphi round, experts indicated the level of priority of each research topic using a 9-point rating scale. Data collection in round two was completed when there was agreement for more than two-thirds of the 21 research topics.

Next, to further scrutinize the pending research questions in depth, qualitative data collection commenced. Using an interview guide, semi-structured telephone interviews, were conducted. Individual interview guides were developed using the individual ratings given to the research topics in the quantitative phase. Interviewees were asked to clarify the priority levels they assigned to certain research topics. Furthermore, interviewees were asked to formulate research questions they would suggest for future studies regarding each of the research topics in the top five. Six interviews, each lasting 30-60 minutes, were conducted audio taped and transcribed verbatim.

3.5 Data analysis

Descriptive statistics of the quantitative data from the two sequential Delphi rounds were obtained using Statistical Package for the Social Sciences 17.0 (SPSS Inc., Chicago, IL). Frequency tables of attributed priority scores were composed and median scores, range of scores and quartiles (Q1-Q3) were calculated. Furthermore, priority scores collected in round two of the Delphi study were analyzed using RIDIT analysis in order to classify the research topics according to an increasing level of priority. RIDIT analysis is a statistically valid method to analyze ordinal data (79,80). In this analysis technique, the distribution of item scores is compared with a reference distribution. We used the distribution of priority scores over all

research topics in our sample of panelists as the reference distribution. The RIDIT obtained for each item reflects the probability that this particular research topic received a higher priority score than another randomly selected topic of the list (81). RIDIT's value ranges from 0 to 1, and higher RIDIT-scores indicate a greater chance that a specific research topic received a higher priority than another randomly selected topic.

Additionally, a Disagreement Index (DI) was calculated for each research topic, to evaluate whether consensus was reached about the assigned level of priority. The DI expresses the diffusion of ratings. To calculate the DI, the Interpercentile Range (IPR) is divided by the Interpercentile Range Adjusted for Symmetry (IPRAS). A DI-value greater than 1 indicates significant disagreement (82).

Finally, qualitative data obtained from the semi-structured interviews were analyzed using a descriptive content analysis, in which verbatim transcripts were labeled.

3. Results

3.1 Quantitative analysis of research priorities in ACHD nursing

Analysis of the priority scores revealed that 17 out of 21 ACHD-related research topics received a median score ≥ 7 on a scale from 1 to 9 (**Figure 2**). The four remaining research topics received a median priority score of five or six. Analysis of the range of scores for each research topic, revealed a great variability in the lowest score, whereas the highest score was always nine, with the exception of one topic that received a maximum score of eight.

Based on RIDIT-analysis, we were able to determine a rank order of research priorities (**Figure 2**). The research topics that were given top priority in ACHD nursing were knowledge and education of patients, followed by outcomes of advanced practice nursing (APN), quality of life, transfer and transition to ACHD care, illness experiences and psychosocial issues,

pregnancy and gynecological issues, health behavior, exercise capacity, role development of APN, organization of care, and palliative care and end-of-life issues. A low priority was given to post-operative pain, sexual functioning, transplantation in ACHD, and health care costs and utilization.

Analysis of the Disagreement Index (DI) demonstrated that there was agreement on the priority scores given to 14 out of 21 (66.7%) suggested topics. There was agreement on the 13 highest ranked research topics, as well as on the lowest ranked topic. There was disagreement among the panel of experts on the priority that should be assigned to the topics regarding long-term survival, survival outcomes of specific interventions, cognition issues, nursing care in general, health care costs and utilization, transplantation in ACHD and sexual functioning.

3.2 Qualitative analysis of research priorities in ACHD nursing

In semi-structured interviews, participants were asked to reflect on specific themes and research questions for the five topics that received the highest priority in the Delphi study. The themes and research questions suggested are listed in **Table 2**. For knowledge and education of patients, specific research questions were formulated with respect to (i) patients' knowledge of the impact of the disease; (ii) development of an educational plan for life; and (iii) learning difficulties or neuro-cognitive impairment in patients with CHD. Research on outcomes of the APN role should specifically target the implementation of different APN roles in ACHD care. For quality-of-life research, it is suggested to undertake studies in specific subgroups of patients with CHD. Furthermore, studies on the development of a transition program for adolescents with CHD, and evaluation of the effectiveness of different models of transitional care are advocated (**Table 2**).

4. Discussion

ACHD is a growing area of practice, one in which a growing number of nurses are directly involved. Numerous ACHD programs have nurse specialists on staff (71) and are assumed to provide evidence-based care. In addition, an increasing number of nurses are undertaking empirical studies in adults with CHD. As providers are preparing for the next decade of ACHD care, it is important to know the priority of topics to be examined in future studies. Therefore, in this study we examined the research priorities as perceived by nurse specialists and nurse researchers in ACHD and, subsequently, developed an international nursing research agenda. The five most important areas for ACHD nursing research were: patient's knowledge and education, APN outcomes, quality of life, transfer and transition to adult CHD care, and illness experiences and psychosocial issues.

Indeed, highest priority was given to studies investigating the knowledge and education of adults with CHD. As pediatric patients move into adult setting, it is assumed they are prepared to become responsible for their life and health care. In order to take up this responsibility, however, adults with CHD need to have sufficient knowledge about their disease, treatment and preventive measures. To date, seven studies were conducted by nurses concerning knowledge and education (48-54). These studies have reported that important gaps in the knowledge of these patients exist concerning their heart defect, treatment and preventive measures; and pointed out that there is a need to develop, implement and evaluate structured educational activities (52,54). In the present study, participants indicated that priority should be given to studies assessing the level of knowledge of patients regarding the need for follow-up, their treatment, pregnancy counseling and physical activities. Further, future research should focus on the development and evaluation of an educational plan for the lifespan of patients.

In our study, the second research priority was research on the outcomes of APN. Indeed, when performing our systematic review, there were no publications identified which investigated the outcomes of the implementation of the APN role. However, to advance the role of ACHD nursing, it is important to measure the impact of APN and to identify targets for improvement. Although during our Delphi study, a lower priority score was assigned to studies examining role development of APN, interviewees stressed however the importance of conducting research on the development and impact of the APN role. Internationally, a number of different roles are defined within the scope of APN. To better define the qualifications, tasks, responsibilities and outcomes of ACHD APNs, future research on role development is needed.

The third priority in the research agenda was quality of life in adults with CHD. To date, ten nurse led studies were conducted on quality of life in ACHD patients (30-38,65). Hence, quality of life is the topic most frequently investigated by nurses involved in ACHD care. Furthermore, a survey among European ACHD nurse specialists showed that 90% of the nurse specialists who were involved in nursing research, conducted quality-of-life research (29). Therefore, it could be counterintuitive that quality-of-life research is still highly needed. The present study, however, revealed that additional studies on quality of life are required, particularly studies conducted in specific subgroups of patients based on for example their age, type of heart defect or other clinical characteristics. Additionally, it is for example, important to identify patients with a high risk for poor quality of life, and investigate interventions to improve their quality of life.

The fourth priority was the transition and transfer of adolescents with CHD to adult-focused facilities. Since the majority of patients with CHD need cardiac follow-up throughout their entire life span (16,18,20,83-85), a timely transfer of care from pediatric cardiology to

different levels of adult-focused care has been recommended (18-20,85). Several consensus documents have described the need for the implementation of transition programs that prepare adolescents with CHD for the transfer of care when becoming an adult (18,27,85). To date, only four nurse-led studies in this area were found. These studies either investigated the experiences and expectations of patients with CHD regarding transfer and transition (56,58), or studied the current practices on transfer and transition in 69 European and North-American centers (55). The need for additional research on the development of transition programs, subsequently followed by experimental studies evaluating the effectiveness of such programs, was expressed in our study.

The fifth research priority was the illness experiences and psychosocial issues of patients with CHD. In order to give patients comprehensive care, attention must be given to both medical and non-medical complications patients might experience (4). Nurses need to comprehend which psychosocial obstacles patients face when growing up with CHD. It is suggested that future research should focus on the impact of illness experiences of adults with CHD in their daily living.

This study was the first to establish an international nursing research agenda for ACHD nursing. However, there are some limitations to bear in mind when interpreting the results of this study. First, only 73% of the panelists who participated in the Delphi study were involved in research. This could imply that nurses gave priority to certain research topics without taking the feasibility of such studies into account. Second, when using the Delphi technique, consideration must be given to the level of consensus to be employed. However, an universally recommended level of agreement does not exist. McKenna et al. suggested a minimal agreement of 51% amongst panelists, while others would recommend to obtain a consensus level of 65%, 70% or

even 80% (63,86). We chose to terminate data collection through the subsequent Delphi rounds, when agreement on the priority scores was obtained for at least two third of the 21 research topics. We calculated a Disagreement Index for each of the 21 predefined research topics in order to evaluate whether agreement was obtained. Third, because we used a mixed methods design, we had the opportunity to investigate the top five research priorities in depth by conducting semi-structured interviews. These interviews revealed that panelists possibly indicated certain research priorities based on their own hospital experience and current working setting suggesting possible bias. For example, when a nurse worked in a hospital where patients were not transferred from pediatric cardiology to an adult-focused facility because the hospital provides one comprehensive program for the entire life-span of the patient, nurses gave a low priority score to this research area.

5. Conclusion

Based on this mixed methods study, an international nursing research agenda on ACHD has been established. Topics that should receive the highest priority are: patient knowledge and education, APN outcomes, quality of life, transfer and transition to adult care, and illness experiences and psychosocial issues. Hence, current and future nurse researchers should consider these areas in order to expand the evidence basis, strengthen the body of knowledge and prepare ACHD nursing for the next decade.

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Figure 1: Flowchart systematic literature review

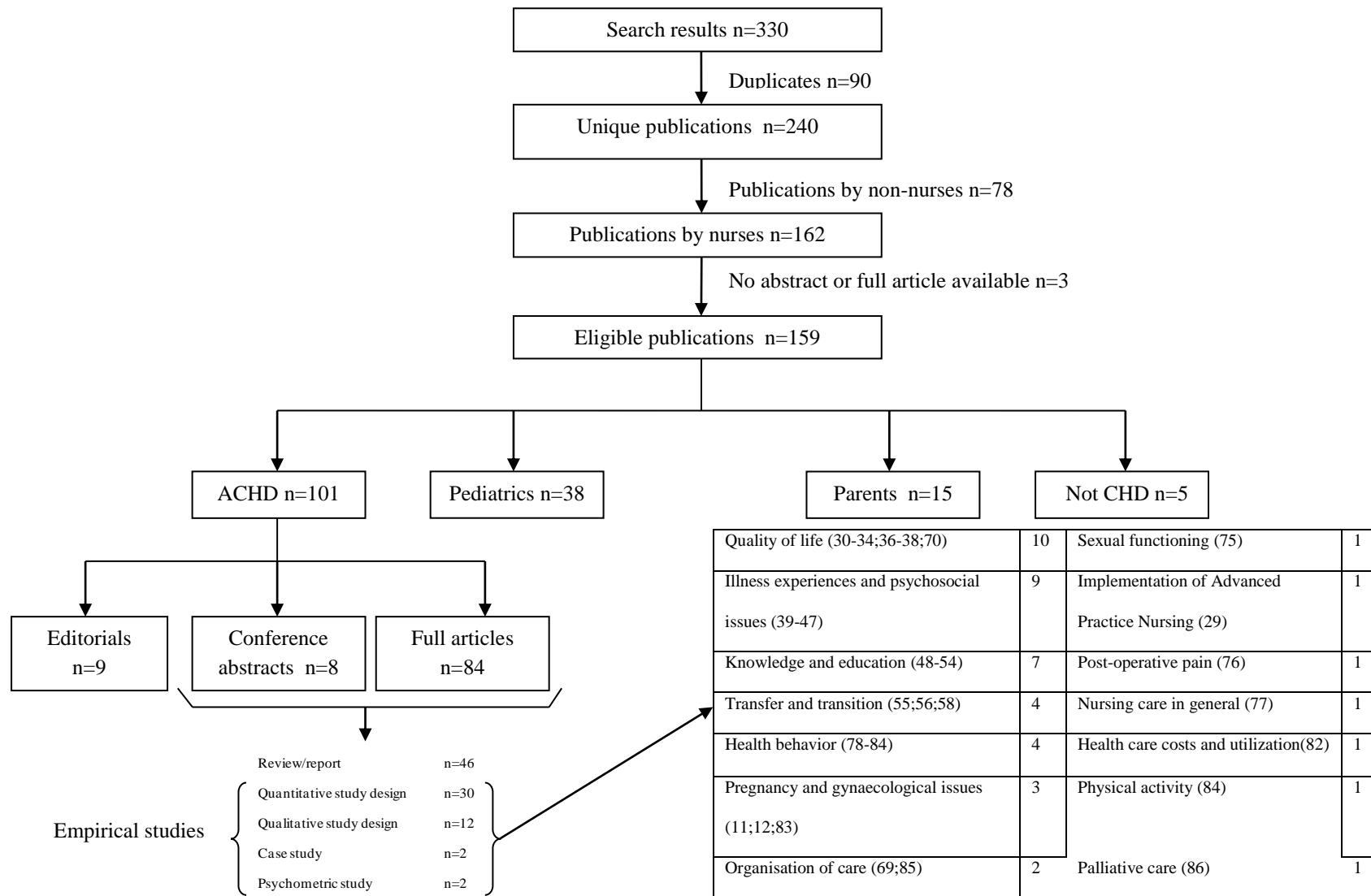
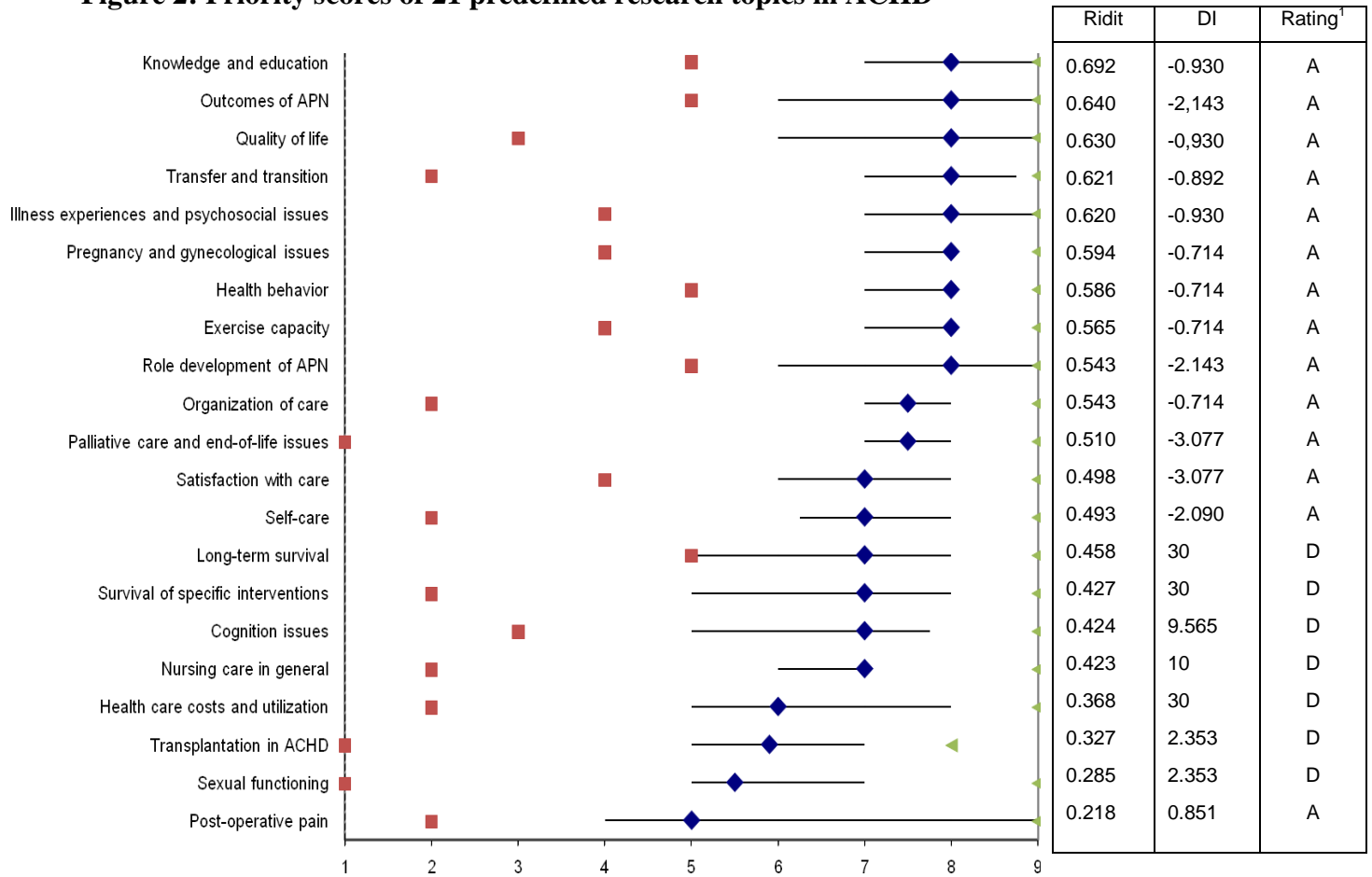


Figure 2: Priority scores of 21 predefined research topics in ACHD



¹A= DI<1, agreement amongst panelists; D=DI>1, disagreement amongst panelists

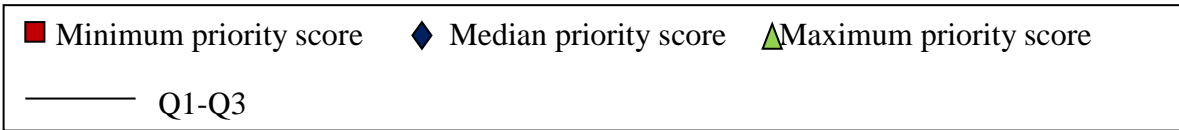


Table 1 Professional characteristics of panelists of the Delphi study(n=37)

Variables	n (%)
Gender	
Female	35 (94.6%)
Male	2 (5.4%)
Age in years (median;Q1-Q3)	44;39-50
Highest level of education	
Certificate in nursing/ Associate degree	5 (13.5%)
Bachelor in nursing/ Polytechnic	4 (10.8%)
Master of Science in Nursing	23 (62.2%)
PhD-student	2 (5.4%)
PhD/Doctorate	3 (8.1%)
Type of hospital	
Local hospital	3 (8.1%)
Regional hospital	1 (2.7%)
University hospital	32 (86.5%)
Other	1 (2.7%)
Work setting (>1 setting possible)	
Outpatient clinic	31 (83.8%)
Cardiac ward/Cardiac surgery	14 (37.8%)
Research/Academic	9 (24.3%)
Education	8 (21.6%)
Other	6 (16.2%)
CCU/ICU	4 (10.8%)
Cathlab	3 (8.1%)
Involvement in nursing research	27 (73.0%)
Years of experience in ACHD (median; Q1-Q3)	5; 3-10
Country	
USA	19 (51.4%)
UK	4 (10.8%)
Canada	4 (10.8%)
Belgium	3 (8.1%)
The Netherlands	3 (8.1%)
Singapore	2 (5.4%)
Denmark	1 (2.7%)
Sweden	1 (2.7%)

Table 2: Suggested research questions on top 5 research priorities in ACHD (n=6)

1. Knowledge and education of patients with CHD

(i) Knowledge on the impact of the disease:

- What is the knowledge of patients on continuity of care (e.g. knowledge on health care insurance, reasoning for follow-up, recommended frequency of follow-up visits, how to navigate through the healthcare system)?
- What is the knowledge of patients regarding pharmacological treatment (e.g. prescribed frequency and doses of medication, possible side-effects, consequences of non-adherence)?
- What is the knowledge of patients, both sexes, on pregnancy counseling (e.g. knowledge on potential risks and complications during pregnancy)?
- What is the knowledge of patients concerning recommended and/or allowed physical activities with regard to their heart condition?

(ii) Development of an educational plan for life:

- What do patients want to know at different stage of their life and their disease?
- Which components from educational models implemented in patients with a chronic condition can be implemented in CHD care?
- Which educational interventions or programs are effective in enhancing the level of knowledge in patients with CHD?

(iii) Learning difficulties and neuro-cognitive impairment:

- What is the proportion of patients with CHD who are neuro-cognitively impaired?
- Which factors could predict the development of learning difficulties in patients with CHD?

2. Outcomes of Advanced Practice Nursing

(i) Evaluation of the implementation of APN roles:

- Which outcomes, parameters and/or indicators should be measured in order to evaluate the impact of different APN roles?
- What is the impact of APN roles in ACHD care on the predefined outcomes, parameters and/or indicators related?

3. Quality of life

- What is the quality of life in the overall sample of patients with CHD?
- What is the quality of life in specific subgroups of patients with CHD (e.g. cyanotic heart lesions) based on their clinical characteristics?
- What is the quality of life in children and adolescents with CHD?
- What is the quality of life in adult patients with CHD?
- What is the quality of life in patients with CHD across different levels of care and settings for cardiac follow-up?
- What are the psychometric properties of the instruments used to investigate quality of life?

4. Transfer and transition to adult health services

(i) Development of a transition program:

- What is the core content or key elements of a transition program for patients with CHD?

(ii) Evaluation of the effectiveness of transition programs:

- Which outcomes, parameters and/or indicators should be measured in order to evaluate the effectiveness of a transition program in patients with CHD?
- Which factors influence continuity of care in adults with CHD?
- What benefits could a transition program give to patients, parents, healthcare workers and the healthcare system in CHD care?
- What is the effectiveness of different organizational models of transition and transfer of care in patients with CHD?

5. Illness experiences and psychosocial issues

- What is the impact of illness experiences of patients with CHD on their daily living?
- Which psychosocial and behavioral problems occur in adolescents with CHD (e.g. anxiety, depression, etc.)?