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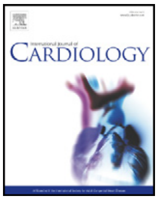

Three parties, one direction: Research priorities in adults with congenital heart disease. What do professionals, patients and relatives want to know?☆

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ABSTRACT

Background: Patients, their relatives, as well as medical specialists from the fields of paediatric cardiology, cardiology and cardiac surgery were surveyed to investigate the current research needs in the field of congenital heart disease (CHD) focussing specifically on the needs of those affected.

Material and methods: An online survey including four groups of patients with CHD (Fontan circulation, transposition of the great arteries [TGA] after atrial switch [AS] and after arterial switch operation [ASO], tetralogy of Fallot [TOF]) was performed. Each questionnaire comprised twelve topics. Persons affected (patients and relatives) were surveyed by means of CHD group specific questionnaires. Participants were recruited through the German National Register for Congenital Heart Defects.

Results: N = 596 affected persons (Fontan circulation: n = 189; TGA after AS: n = 64; TGA after ASO: n = 90; TOF: n = 253) and 75 physicians (57.3% paediatric cardiologists, 28.0% cardiologists, 10.7% cardiac surgeons, 4.0% other) participated. In general, those affected assume a greater need for research than physicians. Regarding the CHD related topics to be the focus of future research, those affected largely agreed with the participating physicians, although with a different ranking of research topics.

Conclusions: Based on the results of our study the challenges immanent in routine care for the CHD patient groups investigated can be identified. Accordingly, these topics should be prioritized in the research of the coming years.

1. Introduction

So far, investigations to identify future research topics in the field of congenital heart disease (CHD) were carried out by means of surveys either among professionals or patients. While most surveys among professionals concentrated exclusively on medical topics, the focus of patient surveys was placed on patients' general desires, worries and needs regarding medical, psychosocial or lifestyle issues [1,2]. The present investigation by means of an online survey aimed at covering both physicians' assessment of the need for future research, as well as capturing the perspective of those affected. For this purpose, patients with CHD, their relatives and medical experts from the field of CHD were surveyed. Assessing the importance assigned to individual research topics from both a medical and the patients' point of view should allow for future research to be specifically geared to the patients' needs without losing sight of the medical facts. Knowing patients', relatives' and physicians' view regarding the importance of different research topics should also help designing targeted studies, the results of which will most probably find their way into clinical routine, while considering aspects of time and cost efficiency [1]. Moreover, the assessment of the

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future need for research sheds light on the implicit need of information regarding medical and psychosocial issues.

The key questions of the study were:

- Are there research topics that are assessed differently by female and male participants?
- Do the surveyed groups (patients, relatives, physicians) differ with respect to their assessment of the future need for research?

2. Material and methods

The National Register for Congenital Heart Defects (NRCHD) is a voluntary register for patients with CHD in Germany. Parents are also given the opportunity to enrol their child with CHD in the NRCHD. After registration the patients and their families have the opportunity to take part in studies, to provide blood and tissue samples to the register's biobank and to receive information on the current state of research in the field of CHD. There is close cooperation between the NRCHD, patient organizations and parents' associations, who promote registration in the NRCHD through their websites and print media. Furthermore, physicians in private practise attending to CHD patients, as well as German heart centres promote registration in the NRCHD. The NRCHD currently comprises data from approx. 49,468 members (as of October, 2015) including children, adolescents, and adults with CHD. This makes it the largest patient database for CHD in Europe.

Four groups of CHD patients that represent a chronically ill population were chosen from the patient collective of the NRCHD (Fontan circulation, transposition of the great arteries [TGA] after atrial switch [AS], TGA after arterial switch operation [ASO], tetralogy of Fallot [TOF]). These patients require continuous and lifelong medical follow-up. The group of “Fontan circulation” is characterized, above all, by the unclear long-term outcome with respect to morbidity and mortality. The two groups of “TGA after AS” and “TGA after ASO” stand out due to the evolution in surgical methods available. While a high burden of morbidity and mortality has already been established for the group of “TGA after AS”, the morbidity and long-term complications to be expected for the group of “TGA after ASO” are as yet largely unknown. Patients assigned to the “TOF”-group often have to face the necessity of reoperations/reinterventions during the course of their disease.

The survey was directed primarily to the patients with CHD. However, our experience in online surveys has shown that often, the patients do not fill out the questionnaires themselves. To prevent a statistical bias, the questionnaire was supplemented by the question as to who had completed the questionnaire, i. e. if it were the patients themselves or one of their relatives. Patients with chromosomal abnormalities such as trisomy 21 are usually not able to fill in complex questionnaires in accordance with the requirements. Therefore, the criteria for inclusion were the presence of a cardiac main diagnosis/cardiac condition qualifying for one of the four CDH groups, the availability of a current e-mail address (of adult individuals) and the absence of chromosomal abnormalities that limit cognitive capacity. Experts in the field of CHD compiled research topics of potentially high clinical significance. On this basis, a list consisting of twelve groups of topics each was compiled for each of the four CHD groups; the compiled sets of questions were divided into two categories (“primarily psychosocial” and “primarily medical”) and reviewed by an expert panel.

The expert panel comprised of (paediatric) cardiologists or congenital cardiac surgeons with long-standing experience in the field of CHD. In addition to representatives of German heart centres, recognized academics in the field of CHD and the members of the Management Boards of the German Competence Network for Congenital Heart Defects (CNCHD) and the NRCHD were included in the expert panel. An overview of the ensuing 17 groups of topics and their classification as “primarily psychosocial” and primarily medical” is given in Table 1.

During the first stage, patients and their relatives, as well as physicians from the fields of paediatric cardiology, cardiology and cardiac surgery were informed about the survey and asked for participation by mail and via the websites of both the NRCHD and the CNCHD. Those affected (patients and relatives) were queried based on respective diagnosis. Over a period of 30 days, the invited participants could access the online questionnaire. Each questionnaire comprised twelve research topics/problem areas that were largely identical in the different CHD group specific versions. Some issues specific to individual CHD groups and weren't asked in the other groups accordingly.

The surveyed physicians were provided with just one single questionnaire consisting of all the four CHD sub-group specific questions (48 questions). This facilitated a comparison of the patients'/relatives' and the physicians' assessment of the need for research.

To prevent multiple entries from the same participant, the computer terminal used for participation was automatically cross-checked against a previous data entry. In addition, each participant was requested to generate a personal code at the beginning. To assess the need for research regarding each topic, participants could choose from a scale of 1 (no need for research) to 6 (great need for research). Those affected were additionally asked for their age, sex, educational background and employment status. From the surveyed physicians, the sex, field of work and function was recorded.

After the 30 day period, during which five different questionnaires were available, the survey was closed and the survey participants were informed of the survey being completed via social media and the online presence of the NRCHD. The collected data were analysed. Approval by the appropriate ethics committee was obtained.

2.1. Statistical analysis

Continuous variables are presented as mean ± standard deviation, while categorical variables are presented in absolute numbers or as percentages. Comparison between groups was performed using a Kruskal–Wallis-test and a Mann–Whitney U test for continuous variables. All tests were performed two-sided and for all analyses, a p-value < 0.05 was considered statistically significant. Statistical analyses were performed using SPSS version 22.

3. Results

3.1. Study participants

Overall, 1104 patients were invited via e-mail to participate in the survey. The participation of 596 patients/relatives corresponds to a response rate of 54% (Fontan circulation $n = 189$; TGA after AS $n = 64$; TGA after ASO $n = 90$; TOF $n = 253$). Table 2 provides an overview over those affected. Additionally, 75 physicians (57.3% paediatric cardiologists, 28% cardiologists, 10.7% cardiac surgeons, 4% other) from all over Germany, Austria and Switzerland participated (Table 3). With respect to average age, the patient group “TGA after ASO” was the youngest CHD group, while the overall proportion of female participants (patients and relatives) was considerably higher than that of the male participants across all groups (with the exception of the group of “TGA after AS”).

3.2. Assessment of the need for research

In ten of the twelve topics, those affected in the CHD group “Fontan circulation” rated the overall need for research to be significantly higher than the group of physicians. In part considerable, but no significant differences were found comparing the assessment of single research areas' importance by female and male patients/relatives (Fig. 1). Regarding six of the twelve topics, the surveyed patients considered the need for research to be significantly lower than their relatives did (Table 4). Regarding seven of the twelve topics, the surveyed patients considered