



Meeting the challenge: The evolving global landscape of adult congenital heart disease



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ABSTRACT

Background: Only limited information is available on the worldwide distribution and volume of adult congenital heart disease (ACHD) centers. We aimed to assess the centers using a bibliometric approach.

Methods and results: We identified publications presenting original research in the field of ACHD between 1995 and 2011. A total of 94,119 articles were identified which underwent electronic filtering and manual review. Overall, a dramatic increase in ACHD publications was seen over the study period. This was accompanied by a matching increase in impact factors and an over-proportional rise in ACHD contributions relative to the general academic field. Research output correlated well with self-reported patient volume and the number of identified ACHD centers in Europe and North America was in agreement with published surveys, thus validating our methodology. We observed a steady increase in the number of publishing ACHD centers worldwide. The number of ACHD centers per 10-million population was highest for Europe (3.6), followed by North America (1.7), Oceania (1.5), South America (0.4), Asia (0.3) and Africa (0.1). In addition, we evaluated the relative research output between developed and emerging economies and provide an overview over the main areas of research in the ACHD field.

Conclusions: Global interest in ACHD is increasing and this is reflected, both, in the number of publishing centers and the volume of research. Our data provides insights into the geographical and temporal distribution of ACHD research over the last 1 1/2 decades. These results could serve as benchmarks for international comparisons and guide efforts for improving ACHD infrastructure.

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1. Introduction

Life expectancy and the quality of life of patients born with a cardiac defect have improved dramatically over the last decades. Over 90% of these patients are now expected to survive to adulthood in high-income countries [1]. In low and middle-income countries, however, survival prospects very likely continue to be compromised. Although accurate global data are lacking, it is anticipated that the number of adults with congenital heart disease is increasing in all countries due to continuing and past global efforts to establish pediatric cardiology and cardiac surgery services.

The need to provide specialized care for adult congenital heart disease (ACHD) patients has been long recognized in Europe and North America [2,3]. This has led to the establishment of an increasing number of ACHD programs [4,5] and has been accompanied by a rising volume of publications trying to close the knowledge gap in this emerging

field [6]. Furthermore, national and international guidelines have been published [2,7], which in turn have contributed to improved outcomes for ACHD patients [8]. Challenges, however, remain even in resource-rich settings. These include adequate implementation of guidelines and research findings into clinical practice, as well as training of an appropriate number of specialists (both medical and surgical) to serve the field [9]. It has also been suggested that the quality of patient care can vary significantly between facilities and may not always meet the complex needs of ACHD patients [3].

To enhance collaboration between ACHD programs, inform junior colleagues seeking training opportunities, guide policy decisions and not least patients in need of ACHD care, surveys have been performed to identify self-declared ACHD centers in Europe and the United States (US) [5,10,11]. Although immensely helpful, the results of these surveys cannot be easily extrapolated to other settings. Furthermore, as longitudinal data is largely lacking – these studies provide little insight into temporal trends. We hypothesized that a bibliometric approach, measuring research output, may be equally feasible, would correlate well with survey data and provide, for the first time, a global perspective and novel information on temporal changes in the ACHD field. This is based on the notion that research is an integral part of ACHD care

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provision, and publications should reflect the work-load of individual centers. While it is theoretically possible that large volume of ACHD programs without publication outputs exist, a substantial patient-base represents a *conditio sine qua non* for achieving high impact publications in the ACHD domain – an area still dominated by clinical research. Furthermore, as this data is refereed and in the public domain it is easy to verify. The data presented here also has policy implications, as it reflects contemporary progress made at national level in improving ACHD activity with time. Furthermore, we identify the main areas of research, including potential temporal trends in research interest, in the field.

2. Methods

We searched PubMed for publications focused on ACHD using a list of keywords consisting of MeSH terms included in the PubMed category “Heart Defects, Congenital”. This list was supplemented with phrases not listed in this category but identified from the tables of content of the current European and American guidelines for ACHD [2,7]. Electronic queries using these terms were performed using PubMed E-utility and R-package version 2.13.0 (R Foundation for Statistical Computing, www.R-project.org). Identified articles underwent electronic filtering, and thereafter manual review. Publications including only pediatric patients were excluded. The process focused on identifying articles written in English and concentrating on original research in the field of ACHD published between 1995 and 2011. All electronically identified articles were subsequently manually reviewed (RFJ and GPD) to exclude non-ACHD publications. We used a three-pronged approach to shed light on global ACHD research and infrastructure: First, we identified cities with at least one publication in the field of ACHD, demonstrating that at least some interest in ACHD exists locally. Although, it could be disputed that this – in itself – is sufficient to presume the existence of a genuine ACHD center, we believe, it represents the most optimistic view of the level of global service provision and should help to place all centers on the map. Secondly, we provide information on the quantity and quality of ACHD research. This should help to identify the global “big players”. Thirdly, we show temporal trends in ACHD over the last 15 years.

Articles were linked to country and city of origin according to the PubMed affiliation field representing the attachment of the first author. Impact factor (IF) data was associated with each publication based on journal and publication year. Journals without IF for a given year were given an IF of zero. IF – data for 2011 were assumed to be identical to 2010. Data on the global population, individual country area and gross domestic product (GDP) were retrieved from the World Bank database (<http://www.worldbank.org/>). The human development index (HDI) was retrieved from the United Nations Development Program (<http://hdr.undp.org/en/statistics/>) and the state fragility index (SFI) 2008 from The Center for Systemic Peace website (www.systemicpeace.org/SFI/matrix08c.pdf). Data relating to ACHD patient and surgical volume for the US were retrieved from the Adult Congenital Heart Association database [11].

2.1. Statistical analysis

Categorical data are presented as frequencies or percentages and comparisons between subgroups were performed using a Chi-square test. The correlation between the number of European centers (according to published survey data) and the number of centers estimated by our approach was assessed using Spearman's rank correlation, while the association between SFI, GDP or HDI and number of ACHD centers per country was evaluated using zero-inflated negative binomial regression methods (to account for the excess of countries with zero ACHD centers) after adjustment for country population. For all analyses, a 2-tailed p-value <0.05 was used as a criterion for statistical significance. R – package version 2.13.0 was employed for all analyses.

3. Results

The initial PubMed search revealed 94,119 articles containing at least one of the keywords from the predefined keyword list. In the electronic filtering process 82,129 publications were excluded, including articles not in English (24,800), those published before 1995 (15,995), articles focused entirely on pediatric cardiology (14,004) or not presenting original research findings (27,330). After manual selection, 4,083 articles were included into the final analysis.

A steep increase in the number of publications and cumulative annual ACHD-IF was evident over time: In 1995, only 129 articles with a cumulative IF of 440 were published, compared to 480 articles with a cumulative IF of 1596 in 2011. This corresponds to an increase of 272% in number and 263% in IF. When compared to the total number of articles listed in

Table 1
Global distribution of ACHD centers by country.

	n	Centers per 10 million population	Centers per 106 km ² area	% world population	
Europe	190	3.6	35.2	7.70%	–
Austria	4	4.8	48.5	0.12%	–
Belgium	6	5.5	198.2	0.16%	–
Czech Republic	2	1.9	25.9	0.15%	–
Denmark	4	7.2	94.3	0.08%	–
Finland	3	5.6	9.9	0.08%	–
France	21	3.2	38.3	0.95%	–
Germany	39	4.8	111.9	1.19%	–
Greece	4	3.5	31.0	0.17%	–
Hungary	2	2.0	22.1	0.15%	–
Iceland	1	31.5	10.0	0.00%	–
Ireland	2	4.5	29.0	0.07%	–
Italy	29	4.8	98.6	0.88%	–
Malta	1	24.2	3125.0	0.01%	–
Netherlands	10	6.0	296.5	0.24%	–
Norway	5	10.0	16.4	0.07%	–
Poland	8	2.1	26.3	0.56%	–
Portugal	7	6.6	76.5	0.16%	–
Romania	1	0.5	4.3	0.31%	●
Slovak Republic	1	1.8	20.8	0.08%	–
Slovenia	2	9.7	99.3	0.03%	–
Spain	8	1.7	16.0	0.67%	–
Sweden	5	5.3	12.2	0.14%	–
Switzerland	5	6.4	125.0	0.11%	–
United Kingdom	20	3.2	82.7	0.91%	–
North America	106	1.7	4.9	8.99%	–
Canada	12	3.5	1.3	0.50%	–
USA	90	2.9	9.8	4.52%	–
Guatemala	1	0.7	9.3	0.21%	●
Mexico	3	0.3	1.5	1.66%	●
Oceania	6	1.5	0.7	0.59%	–
Australia	5	2.2	0.7	0.33%	–
New Zealand	1	2.3	3.8	0.06%	–
South America	14	0.4	0.8	5.73%	●
Argentina	1	0.2	0.4	0.59%	●
Brazil	9	0.5	1.1	2.85%	●
Chile	1	0.6	1.3	0.25%	●
Colombia	2	0.4	1.8	0.68%	●
Uruguay	1	3.0	5.7	0.05%	–
Asia	129	0.3	2.7	61.72%	●
China	28	0.2	3.0	19.56%	●
Georgia	1	2.2	14.4	0.07%	–
India	16	0.1	5.4	17.12%	●
Indonesia	1	0.0	0.6	3.51%	●
Iran	4	0.2	2.2	3.51%	●
Israel	7	9.2	323.5	0.11%	–
Japan	43	3.4	118.0	1.86%	–
Korea, Rep.	5	1.0	51.5	0.71%	–
Lebanon	1	2.4	97.8	0.06%	–
Malaysia	2	0.7	6.1	0.42%	●
Pakistan	2	0.1	2.6	2.54%	●
Philippines	1	0.1	3.4	1.36%	●
Russian Fed.	2	0.1	0.1	2.07%	●
Saudi Arabia	1	0.4	0.5	0.40%	●
Singapore	1	2.0	1428.6	0.07%	–
Thailand	4	0.6	7.8	1.01%	●
Turkey	10	1.4	13.0	1.06%	–
Africa	9	0.1	0.3	15.04%	●
Algeria	1	0.3	0.4	0.52%	●
Cameroon	1	0.5	2.1	0.29%	●
Egypt	1	0.1	1.0	1.19%	●
Ghana	1	0.4	4.4	0.36%	●
Kenya	1	0.2	1.8	0.59%	●
Nigeria	2	0.1	2.2	2.32%	●
Sudan	1	0.2	0.4	0.64%	●
Tunisia	1	0.9	6.4	0.15%	●

Countries without scientific output in the field are not listed. These countries, by definition, have zero ACHD centers per 10 million population. Russian Fed.—without accounting separately for European and Asian part. ● = country with less than one center per 10 million population.

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