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REVIEW ARTICLE

Congenital diaphragmatic hernia: a scientometric analysis of the global research activity and collaborative networks

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Abstract

Despite a growing interest to clinicians and scientists, there is no comprehensive study that examines the global research activity on congenital diaphragmatic hernia (CDH). A search strategy for the Web of Science™ database was designed to identify scientific CDH publications. Research output of countries, institutions, individual authors, and collaborative networks was analyzed. Semi-qualitative research measures including citation rate and *h*-index were assessed. Choropleth mapping and network diagrams were employed to visualize results. A total of 3669 publications were found, originating from 76 countries. The largest number was published by the USA ($n=1250$), the UK ($n=279$), and Canada ($n=215$). The USA combined the highest number of cooperation articles ($n=152$), followed by Belgium ($n=115$) and the Netherlands ($n=93$). The most productive collaborative networks were established between UK/Belgium ($n=53$), Belgium/Spain ($n=47$), and UK/Spain ($n=34$). Canadian publications received the highest average citation rate (22.8), whereas the USA had the highest country-specific *h*-index (72). Eighty-five (2.3%) articles were published by international multicenter consortiums and national research networks. The most productive institutions and authors were based in North America and Europe. Over the past decades, CDH research has increasingly become multidisciplinary and numerous innovative therapeutic strategies were introduced. CDH-related research has constantly been progressing, involving today many disciplines with main research endeavors concentrating in a few high-income countries. Recent advances in prenatal interventions and regenerative medicine therapy hold the promise of improving CDH outcome in the 21st century. International collaborations and translational research should be strengthened to allow further evolution in this field.

Keywords Congenital diaphragmatic hernia · Pulmonary hypoplasia · Pulmonary hypertension · ECMO · FETO · Bibliometrics

Introduction

Congenital diaphragmatic hernia (CDH) is a life-threatening defect in the integrity of the developing diaphragm, which accounts for approximately 8% of all major congenital malformations [1]. In the United States and Europe, current incidence rates range between 1.9 and 2.3 per 10,000 births [2, 3]. Although numerous chromosomal aberrations and gene mutations have been linked to CDH, the etiology of the diaphragmatic defect is identified in less than 50% of patients [4]. The opening in the diaphragm allows abdominal organs to herniate into the thoracic cavity, thereby competing for space that would normally accommodate the growing lungs [5, 6]. As a result, lung development is severely affected, leading almost invariably to pulmonary hypoplasia and persistent pulmonary hypertension of the newborn [7]. Depending on the extent of this unfortunate combination, neonates with CDH frequently suffer from respiratory distress at birth, which can be lethal in up to 40% of cases [8, 9]. Despite significant advances in postnatal resuscitation and use of lung-protective treatment strategies, CDH remains one of the major therapeutic challenges in modern neonatal intensive care, causing high mortality and serious long-term morbidity for survivors [10, 11].

As CDH continues to be a relatively complex and rare birth defect with often unpredictable outcome, there is urgent need to foster further research activities in this field, which will mainly depend on multi-institutional and

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international collaborations [12]. An appreciation of CDH literature and scientific progress is, therefore, essential for both clinicians and basic scientists to plan future research projects. However, due to the enormous quantity and heterogeneity of CDH-related publications, it is difficult for a single researcher to survey all the published items to gauge their individual scientific value, and thus to stay abreast of the latest research evidence.

Scientometrics is an emerging field that studies quantitative aspects and characteristics of scientific research [13]. The main areas of scientometric analysis focus on the measurement of research productivity and citation impact within the scientific community [14]. In recent years, stakeholders and grant authorities have increasingly been using scientometric benchmarks to assess research performance when ranking applicants for academic appointments or determining eligibility for funding [13, 14]. Until now, no detailed study has systematically analyzed the immense number of publications relating to CDH research and the true extent of the scientific output in this field remains unclear. Hence, the aim of this study was to critically evaluate the global CDH research activity, which, in turn, may help to establish future collaborations and thus to advance patient care. To ascertain countries, institutions, and individual researchers that have produced most CDH work, a combination of validated scientometric methods [15] and novel visualization techniques was used, outlining the intricate architecture of international collaborative networks.

Materials and methods

Information source and search terms

A comprehensive search strategy was designed for the Web of Science™ database (Clarivate Analytics, Boston, USA) to identify all peer-reviewed scientific publications relating to CDH research. This online subscription-based research platform, which provides temporal coverage from the year 1900 to present, was accessed on 20 June 2017. The following linked search terms were used, taking into account alternative nomenclature for CDH: "congenital* diaphragm* hernia*" OR "congenital* diaphragm* defect*" OR "fetal* diaphragm* hernia" OR "pediatric* diaphragm* hernia*" OR "foetal* diaphragm* hernia" OR "paediatric* diaphragm* hernia*" OR "agenes* diaphragm*" OR "agenes* hemidiaphragm*" OR "Bochdalek* hernia*" OR "Morgagni* hernia*". To determine only the most relevant research items, a "title" rather than "topic" search approach was chosen. No language restrictions were imposed. Results from 2017 were excluded from the search to ensure complete data acquisition, because the incorporation of several parameters into the database requires a certain time. Any ambiguities during this search process were resolved by consensus of all authors.

Selection categories and data analysis

The retrieved data on CDH-related publications were critically evaluated and classified with regard to subject category, document type, journal title, publication date, and language. Total research output of countries, institutions, individual authors, and collaborative networks was determined and systematically analyzed. The "*citation report*" function was applied to assess semi-qualitative research aspects including citation rate and *h*-index. The *h*-index is an established metric, which incorporates a citation index and the overall scientific output of authors or institutions, thus quantifying importance, impact, and significance of individual research contributions [16]. It can be calculated, if *h* of all publications received at least *h* citations each. In this study, the *h*-index has also been used to estimate to productivity of the publishing countries. The relationship of two or more authors from different countries, who contributed to a joint publication, was defined as a cooperation article. Complete bibliographic data were downloaded as plain text files and extracted into an electronic datasheet in a standardized manner. Choropleth mapping (i.e., differences in color values to represent geographical data) and network diagrams were employed to visualize results.

Results

Global research productivity

A total of 3669 publications on CDH were identified, originating from 76 countries (Fig. 1). North America and Europe constituted the two scientific centers in the field of CDH-related research. In contrast, the vast majority of African countries had an extremely low or no scientific output on CDH. Globally, the largest number of scientific articles relating to CDH was published by the USA [$n = 1250$; (34.1%)], the UK [$n = 279$; (7.6%)], and Canada [$n = 215$; (5.9%)]. Most CDH papers were written in English [$n = 3432$; (93.5%)], followed by French [$n = 87$; (2.4%)] and German [$n = 81$; (2.2%)].

International research collaborations

Clinicians and basic scientists in 53 (69.7%) of the identified 76 countries that published CDH-related work were involved in international research collaborations (Fig. 2). The USA combined the highest number of cooperation articles on CDH ($n = 152$), followed by Belgium ($n = 115$) and the Netherlands ($n = 93$). The most productive collaborative network

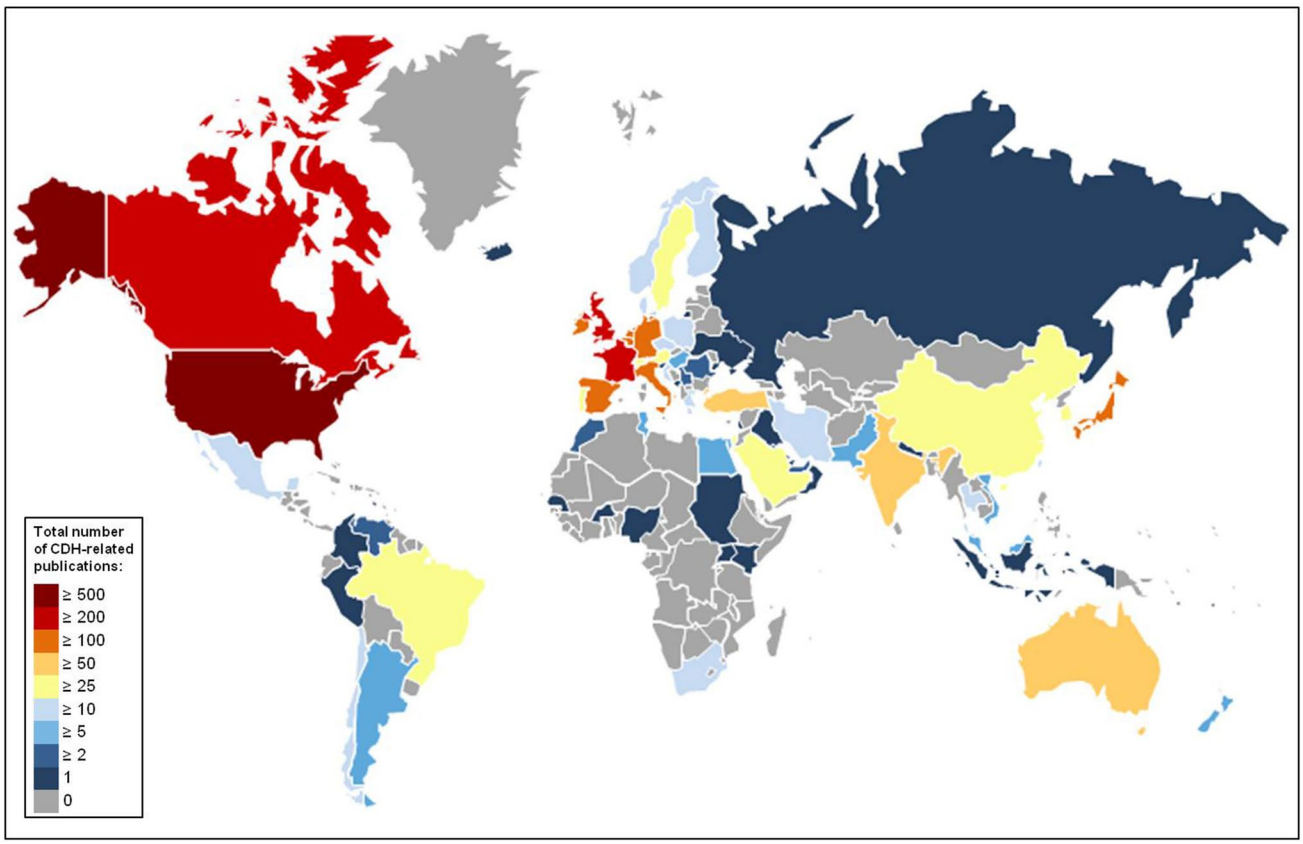

Fig. 1 Choropleth mapping visualizing global publication volume in the field of CDH research

in the field of CDH research was established between UK/Belgium ($n=53$), followed by Belgium/Spain ($n=47$) and UK/Spain ($n=34$). Luxembourg ($n=3$), Venezuela ($n=2$), Dominica, Iceland, Indonesia, Malta, Peru, St. Kitts & Nevis, Sudan, and Ukraine ($n=1$/each) only had joint CDH papers, whereas Turkey had with 3/92 (3.3%) the smallest percentage of cooperative items in relation to its total output. CDH researchers in 23 (30.3%) countries were not involved in any international collaborations. Of those, South Korean investigators released the largest number of CDH publications ($n=28$), followed by authors from Iran ($n=11$) and Tunisia ($n=8$).

Eighty-five (2.3%) scientific articles on CDH were published under the auspices of international multicenter consortiums and national research networks, which addressed various issues associated with this life-threatening birth defect. Table 1 lists the ten most productive CDH cooperations and registries worldwide.

Citation rate and country-specific *h*-index

The 215 identified CDH publications from Canada had the highest average citation rate per published item (22.8), followed by articles from the Netherlands (20.7) and USA (20.2). The USA had the highest country-specific *h*-index in the field of CDH-related research (72), followed by Canada (40) and the UK (38). In contrast, many scientific papers from African, Middle Eastern, and Eastern European countries received extremely few citations, and thus, these countries frequently had an *h*-index of 1 or 0.

Most productive research institutions and authors

All 3669 scientific publications on CDH were evaluated in relation to their institutions of origin and contributing authors. The identified CDH articles were affiliated with a total of 2187 institutions and 10,210 authors. The ten most productive CDH research institutions in the world were located in the USA, the Netherlands, Belgium, France, Ireland, the UK, and Canada (Fig. 3a). The ten most productive authors (appearing anywhere in the author list) in the field of CDH-related research came from the USA, Belgium, the Netherlands, Ireland, Spain, and Germany (Fig. 3b).

Scientific subject categories and document types

Subject categories are defined standard categories in the Web of Science™ database, which represent general areas

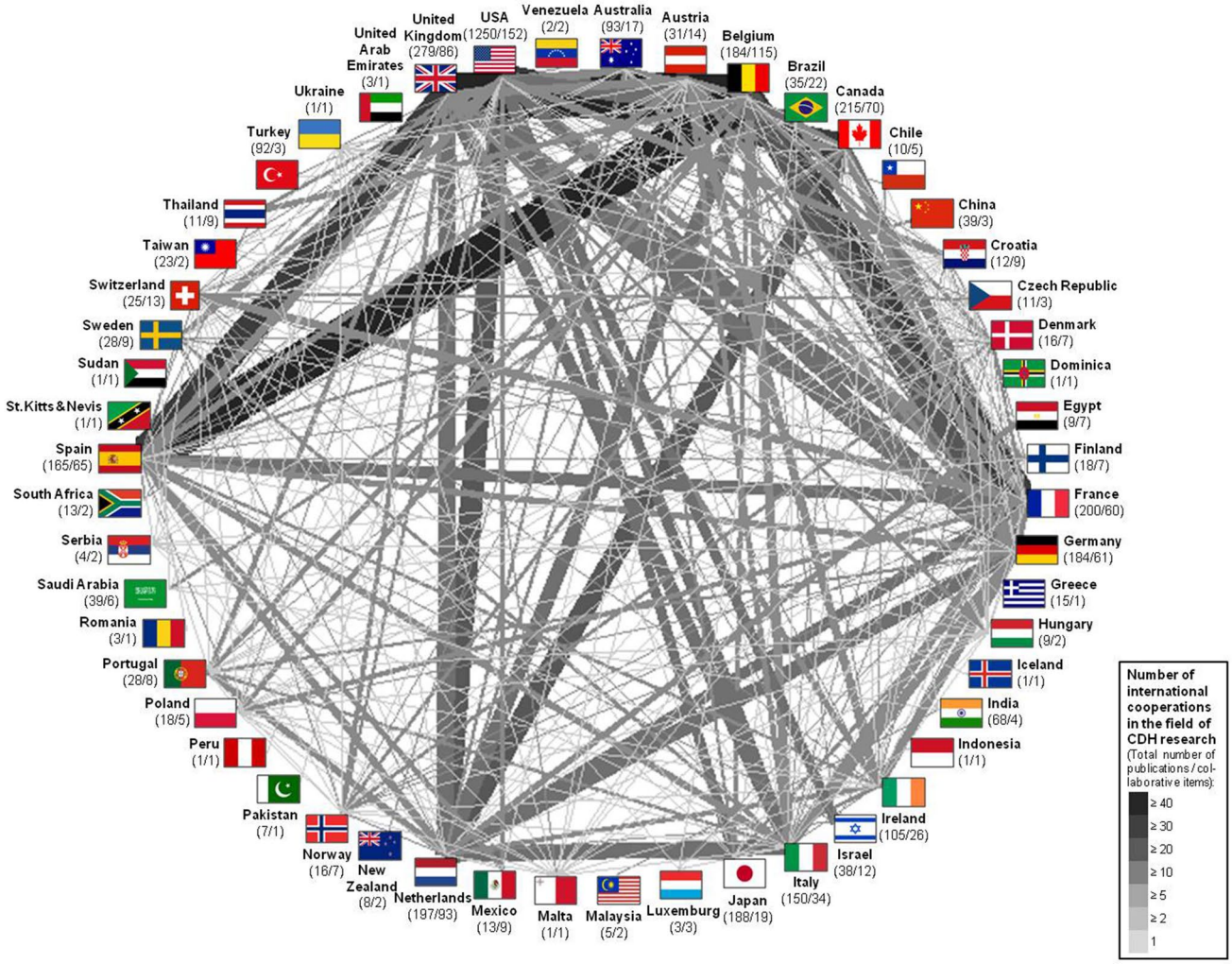

Fig. 2 Network diagram of international collaborations and cooperation publications relating to CDH

Table 1 Ten most productive national and international CDH cooperations and registries

Rank	Name of cooperation or registry	Year of foundation	Participating centers	Extent of collaboration	Publications
1	CDH Study Group (CDHSG)	1995	$n=112$	14 countries worldwide	$n=35$
2	Canadian Pediatric Surgery Network (CAPSNet)	2005	$n=17$	Canada nationwide	$n=14$
3	FETO Task Group	2004	$n=5$	4 European countries, 1 U.S. center	$n=6$
4	CDH EURO Consortium	2008	$n=22$	13 European countries, 1 Canadian center	$n=5$
5	French CDH Study Group/Center for Rare Diseases—CDH	2007	$n=31$	France nationwide	$n=5$
6	Japanese CDH Study Group	2011	$n=9$	Japan nationwide	$n=5$
7	Antenatal CDH Registry Group	2005	$n=10$	5 European countries, 1 U.S. & 1 Israeli center	$n=3$
8	Groupe Radiopédiatrique de Recherche en Imagerie foetale (GRRIF)	1980	$n=10$	2 European countries	$n=2$
9	National Birth Defects Prevention Study (NBDPS)	1996	NS	10 U.S. states	$n=2$
10	Western Canadian ECMO Follow-Up Group	1997	$n=5$	Western Canada	$n=2$

NS not stated

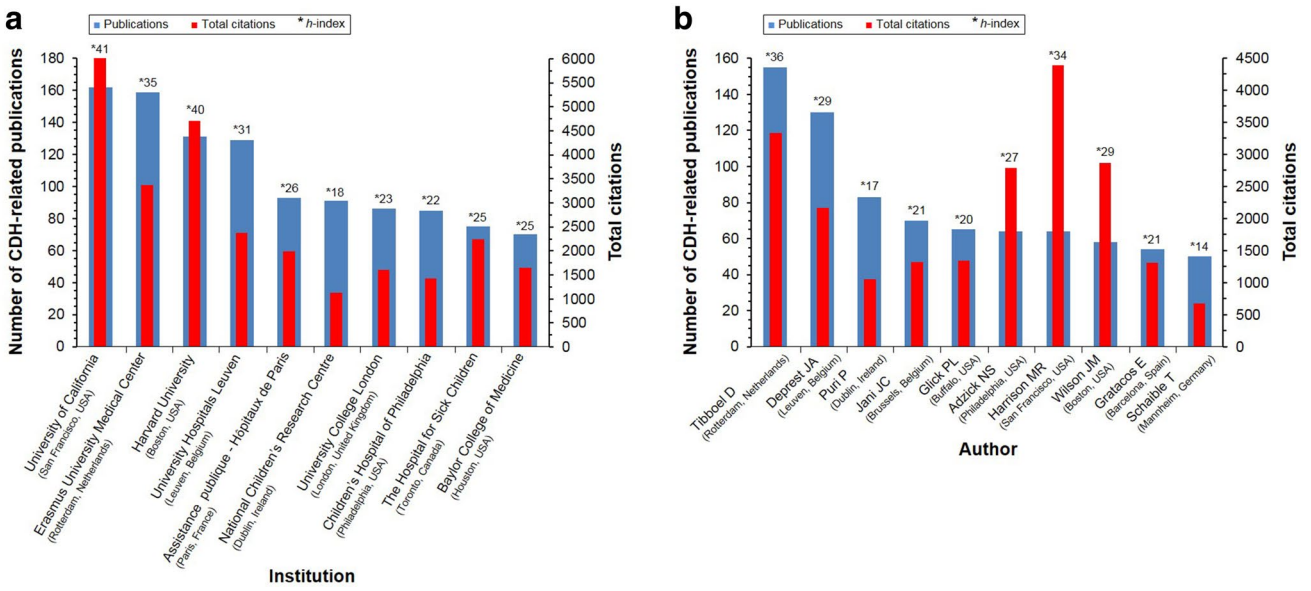

Fig. 3 Ten most productive institutions (**a**) and authors (**b**) in the field of CDH-related research

of science. These categories were distributed by the Journal Citation Reports™ for each scientific journal and its publications. Most articles relating to CDH research were assigned to the subject category "PEDIATRICS" ($n = 1723$), followed by "SURGERY" ($n = 1474$) and "OBSTETRICS/GYNECOLOGY" ($n = 449$). Other common categories were "GENERAL INTERNAL MEDICINE" ($n = 370$), "RADIOLOGY/NUCLEAR MEDICINE/MEDICAL IMAGING" ($n = 259$), "RESPIRATORY SYSTEM" ($n = 212$), "GENETICS" ($n = 188$), "CARDIOVASCULAR SYSTEM/CARDIOLOGY" ($n = 96$), "RESEARCH/EXPERIMENTAL MEDICINE" ($n = 74$), and "GASTROENTEROLOGY/HEPATOLOGY" ($n = 66$). Document types of the 3669 identified CDH publications were classified as 2576 original articles (70.2%), 494 meeting abstracts and proceedings (13.5%), 332 editorials and letters (9.0%), 149 reviews (4.1%), and 118 others (3.2%).

Publication and citation trend

The first CDH-related paper was published in 1910 and the number of subsequent scientific publications increased almost annually, associated with a steady increase in citations (Fig. 4). Until 1970, there was low publication activity, comprising of 161 articles. From 1970 onwards, the number of published items increased constantly with a steep rise in the early–mid 1990s, interrupted by a brief drop in the late 1990s/early 2000s, comprising a total of 3508 articles (i.e., 95.6% of all the scientific publications on CDH were published after 1970). Overall, authors published 19-fold more articles relating to CDH in 2016 than in 1970. Between 1922 and 2016, the 3669 identified CDH publications received a total of 51,253 citations and an average of 533.9 citations per year (range, 0–3215).

Notable scientific journals and publications

All scientific journals listed in the Web of Science™ database were examined in regard to their individual output relating to CDH research and citation rates of relevant items were determined. The 3669 CDH-related articles were published in 573 different journals with an average citation rate of 14.0 (range 0–414) per publication (*h*-index 85). The "*Journal of Pediatric Surgery*" was identified as the most productive journal (n = 649), whereas "*The Journal of Pediatrics*" had with 33.1, the highest average citation rate per published CDH paper (Fig. 5).

Table 2 lists the ten most-cited articles in the field of CDH research during this time span.

Discussion

The present study draws the first detailed map of global CDH research architecture based on an in-depth analysis of the scientific output from 1910 to 2016. During this time span, a total of 3669 publications on CDH were indexed in Web of Science™ database, originating from 76 countries. A small number of North American and European countries were responsible for the majority of CDH-related research, which not only generated most of the scientific articles, but also papers high in quality. Of these, the USA, Canada,

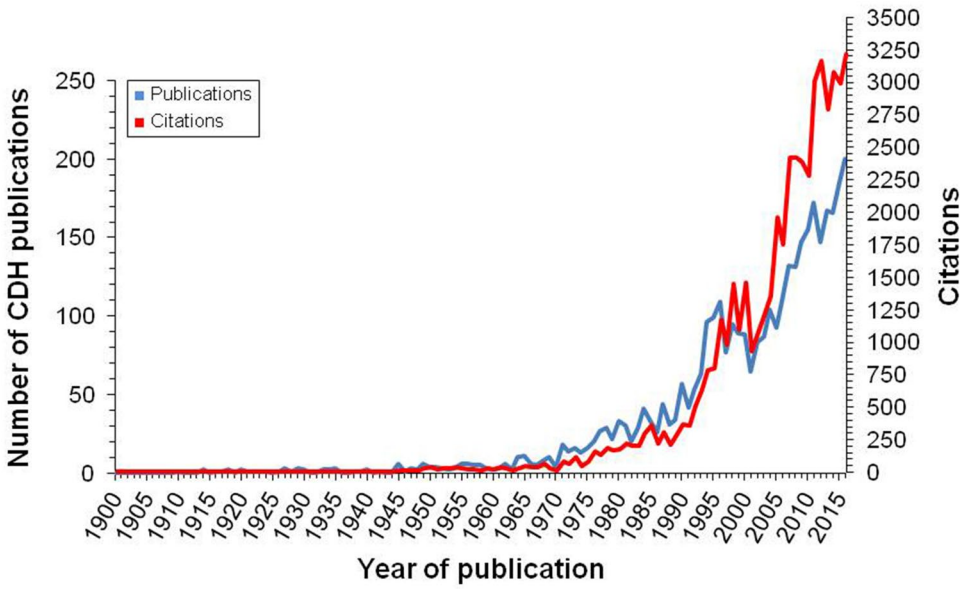

Fig. 4 Overall number of CDH publications and received citations in the time span 1900–2016

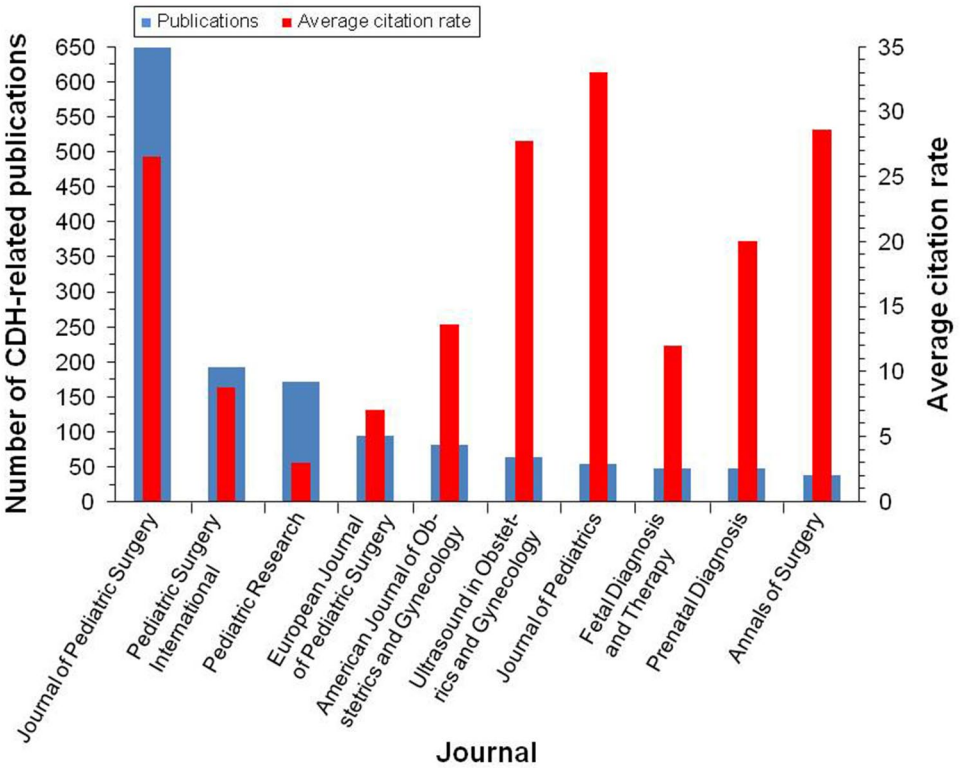

Fig. 5 Ten most productive journals with regard to CDH publications and average citation rate per published article

the UK, France, and the Netherlands were the five leading countries with regard to the total number of CDH publications, average citation rate, and *h*-index. This mirrors the worldwide trend for a greater volume of scientific articles to originate from high-income countries [17, 18], and furthermore, for authors from these countries to dominate key roles in authorship. In comparison, the lack of publications from low- and middle-income countries reflects a pattern in all fields of medicine: that survival of infants with conditions such as CDH is often not feasible in countries with low resources or in healthcare systems, where medical professionals are too busy with clinical pressures to commit time to research. As a significant progress cannot be made by a single researcher, there is currently a global movement in science towards strategically designed national or international collaborations to improve overall patient care [19].

Table 2 Ten most-cited publications in the field of CDH research

Rank	Publication	Total citations	Citations per year
1	Cantrell JR, Haller JA, Ravitch MM (1958) A syndrome of congenital defects involving the abdominal wall, sternum, diaphragm, pericardium, and heart. Surg Gyn Obstet 107:602–614	414	6.90
2	Metkus AP, Filly RA, Stringer MD, Harrison MR, Adzick NS (1996) Sonographic predictors of survival in fetal diaphragmatic hernia. J Pediatr Surg 31:148–152	351	15.95
3	Harrison MR, Keller RL, Hawgood SB, Kitterman JA, Sandberg PL, Farmer DL et al (2003) A randomized trial of fetal endoscopic tracheal occlusion for severe fetal congenital diaphragmatic hernia. N Engl J Med 349:1916–1924	300	20.00
4	Stege G, Fenton A, Jaffray B (2003) Nihilism in the 1990s: the true mortality of congenital diaphragmatic hernia. Pediatrics 112:532–535	269	17.93
5	Kitagawa M, Hislop A, Boyden EA, Reid L (1971) Lung hypoplasia in congenital diaphragmatic hernia. A quantitative study of airway, artery, and alveolar development. Br J Surg 58:342–346	244	5.19
6	Boloker J, Bateman DA, Wung JT, Stolar CJ (2002) Congenital diaphragmatic hernia in 120 infants treated consecutively with permissive hypercapnea/spontaneous respiration/elective repair. J Pediatr Surg 37:357–365	224	14.00
7	Harrison MR, Jester JA, Ross NA (1980) Correction of congenital diaphragmatic hernia in-utero – 1. The model—intra-thoracic balloon produces fatal pulmonary hypoplasia. Surgery 88:174–182	210	5.53
8	Difiore JW, Fauza DO, Slavin R, Peters CA, Fackler JC, Wilson JM (1994) Experimental fetal tracheal ligation reverses the structural and physiological-effects of pulmonary hypoplasia in congenital diaphragmatic hernia. J Pediatr Surg 29:248–257	209	8.71
9	Lipshutz GS, Albanese CT, Fekdstein VA, Jennings RW, Housley HT, Beech R et al (1997) Prospective analysis of lung-to-head ratio predicts survival for patients with prenatally diagnosed congenital diaphragmatic hernia. J Pediatr Surg 32:1634–1636	204	9.71
10	Deprest J, Gratacos E, Nicolaides KH; FETO Task Group (2004) Fetal tracheal occlusion (FETO) for severe congenital diaphragmatic hernia: evolution of a technique and preliminary results. Ultrasound Obstet Gynecol 24:121–126	201	14.36

This is particularly relevant for rare conditions like CDH as shown by the increasing number of cooperation papers and collaborative networks in this field. The USA was the most favored cooperation partner with collaborations in 31 countries and 152 joint CDH publications, despite the fact that the percentage of collaborative articles was with 12.2% relatively low. However, the most productive collaborative network on CDH was established between the UK and Belgium. These findings can partly be explained by the efficient and well-funded academic structure in both countries, allowing leading experts in their respective scientific fields more frequently to cooperate with their international colleagues [20, 21]. All of the most productive institutions and authors were either based in North America or Europe.

The absolute number of CDH papers has increased 19-fold since the 1970s, associated with an equally steep increase of citations and replicating the same trend as shown in previous scientometric studies on other pediatric conditions [22]. Advances in postnatal resuscitation and introduction of new therapeutic strategies in the 1990s and 2000s, respectively, most likely contributed to the steep increase of CDH research in these two decades. With over 200 CDH-related publications, the year 2016 was the most productive year ever. Besides a growing scientific interest in CDH worldwide, this development may reflect the rapidly increasing volume of biomedical articles in general and increasing pressure for clinicians and scientists to publish, also known as the "*publish or perish*" climate [23]. Unfortunately, modern academia has created an artificial necessity of publishing, not for the advance of knowledge, but for the advance of professional careers [24]. Furthermore, our analysis of subject categories and scientific journals showed an increasing diversification into medical subspecialties and newer research areas, which also applies for the professional fields that CDH authors currently work in.

International multicenter consortiums and national research networks have addressed many critical knowledge gaps pertaining to CDH care. Most importantly, they have identified variability in both CDH practice and outcome among participating centers. Using combined data from these groups, national or international consensus guidelines for multidisciplinary CDH treatment may be produced to standardize the best practices for patients with CDH, from prenatal diagnosis to hospital discharge, based on the best available clinical evidence. In addition, collaborations with global initiatives such as CDH International [25] may help to foster further research activities and strengthen support groups.

What have been the topics of the most-cited CDH work so far? Four out of the ten most-cited articles were directly linked with the intriguing concept of in-utero intervention for fetuses with CDH, reporting pioneering work from its

experimental beginnings, subsequent evolution of this technique, and a randomized-controlled trial. Although there is currently insufficient evidence to recommend fetal endoscopic tracheal occlusion (FETO) as a part of routine clinical practice [26], a few specialized fetal medicine centers in Europe, and North and South America successfully perform this procedure [27–31]. Recently, it has been reported that FETO improves neonatal survival in CDH fetuses with severe pulmonary hypoplasia compared with the standard perinatal management [32, 33]. Today, FETO results in a survival rate of 50–60% [34]. Further results from ongoing international randomized trials are anticipated in the near future [35]. Two further papers dealt with prenatal predictors for postnatal CDH survival. With the advent of routine maternal ultrasound scanning, CDH can now be diagnosed prenatally in up to 60% of cases [36]. Nowadays, the observed-to-expected lung area-to-head circumference ratio measured on 2D ultrasonography is routinely used by fetal medicine centers around the world as a good indicator of neonatal prognosis and chronic lung disease in survivors with CDH [37, 38]. Other valuable prognostic parameters are extent of liver herniation and observed-to-expected fetal lung volume on magnetic resonance imaging [39, 40]. However, gestational age at diagnosis should be taken into account when estimating postnatal morbidity and mortality [41]. Other highly cited themes were: CDH-associated mortality, pulmonary hypoplasia, and lung-protective therapies. During the last 2 decades, CDH survival rates have slightly but significantly improved [42]. Whereas some specialized centers have reported survival rates of close to 90%, pooled results from the CDH Study Group indicated that today's overall survival rate is approximately 70% [43]. Defective lung alveolarization appears to be a common and potentially actionable phenotype in both patients and animal models of CDH [44]. These findings have revealed opportunities for the development of novel targeted treatment options, particularly in the postnatal stages, when therapeutic drugs combined with appropriate ventilation strategies and extracorporeal membrane oxygenation can have maximum clinical impact on surviving patients. Interestingly, the most-cited CDH publication first described the pentalogy of Cantrell, where the diaphragmatic defect is merely one component of this rare syndrome.

Numerous articles of improved survival rates for cases of CDH patients have prevailed in the literature over the past 20 years, which was often attributed to advances in medical management in the postnatal period. However, Stege et al. [45] reported that the mortality of CDH, when complete case ascertainment is achieved, remains largely unaffected by new therapies. They found that the CDH survival rate is principally determined by the rate of prenatal termination and the incidence of associated anomalies. In addition, recent population-based studies revealed that the reported increase in survival outcomes, which are often single institution-based reports, is confounded by case-selection bias which fails to consider those CDH patients who do not reach the referral centers [46]. This apparent discrepancy between population-based and institution-based statistics raises the question of 'hidden mortality' and the role which it plays in both research and clinical medicine. A hidden mortality certainly exists for institutionally reported CDH survival rates. Therefore, articles of improved survival of CDH should be interpreted with caution, as variations in outcome are more likely to be explained by case-selection artifact. More comprehensive population-based tools are urgently needed for reliable counseling and evaluation of current and future CDH treatments. Increased survival has been accompanied by an increase in neurological, nutritional, and musculoskeletal morbidity among the long-term CDH survivors. Today, nearly 75% of infants with CDH are discharged with one or more major comorbidity, including severe gastrointestinal, pulmonary, and neurological issues, in 61.7%, 30.2%, and 20.4% respectively [47]. Due to the increasing complexity of CDH patient care and associated long-term sequelae, there is a growing trend of multidisciplinary follow-up [48–50].

The World Health Organization has set up with major publishers the health internetwork access to research initiative—HINARI [51], which enables academic researchers and practicing physicians in low- and middle-income countries to gain access to one of the world's largest collections of biomedical and health literature. Similarly, the Research-4Life program [52] provides free or very low-cost online access to the major journals in biomedical and related health sciences to local, not-for-profit institutions in developing countries. As a unique public–private partnership between United Nations agencies, universities, and publishers, it aims to reduce the knowledge gap between developing and industrialized countries, and in turn, contributing to improve world health.

One of the main limitations of this study was related to the search engine used. Although the Web of Science™ database is a well-established platform in citation analysis and one of the most comprehensive, accurate, and unbiased resources for literature searching, not all journals, institutions, or individual authors that published CDH-related research are necessarily listed. The use of other search engines such as PubMed/MEDLINE would likely have resulted in marginally different figures. Additionally, the choice of database may have caused a potential language bias towards scientific articles from English-speaking countries [53], and it is also known that authors and reviewers tend to be biased towards their native language in their citation practice [54, 55]. As the applied search strategy was based on a title rather than a topic search to identify all peer-reviewed papers, which focused primarily on CDH research, a few relevant research items

may not have been recognized by the automated computer search. Another possible bias may be the analysis of citation frequency and *h*-index as measures of scientific quality rather than using journal impact factors as a surrogate [56]. In turn, it must be considered that self-citation by authors can considerably manipulate the *h*-index. Nevertheless, this metric is a proven tool to compare different countries, institutions and authors working in one specific field [57]. Egghe's *g*-index may be a useful alternative to the *h*-index as it aims to improve on the *h*-index by giving more weight to highly cited articles [58]. This metric was first introduced in 2006 as an improvement of the *h*-index of Hirsch to measure the global citation performance of a set of articles. If this set is ranked in decreasing order of the number of citations that they received, the *g*-index is the (unique) largest number, such that the top *g* articles received (together) at least g^2 citations. More recently, Bartneck and Kokkelmans proposed the *q*-index as an indicator for how strategically an author has placed self-citations and thus serves as a tool to detect possible manipulation of the *h*-index [57]. Unfortunately, the Web of Science™ database does not provide raw data in a form, which would allow calculation of these two metrics. One final point to consider is the phenomenon known as the "*Matthew Effect*", where the more cited papers, scientists, and journals get even more cited, the less cited ones get less cited [59]. It has often been assumed that CDH papers, which have been published in journals that only comprise a very small number of CDH-related articles in relation to their overall publication volume, may have less competition within the respective journal and may, therefore, be more frequently cited. However, we believe that the true cause for this slightly distorting citation phenomenon actually lies in the wider scientific coverage of these journals, thus attracting are larger readership, and therefore, these publications are probably more likely to be cited.

In conclusion, this study provides a useful guide for clinicians, scientists, and stakeholders to identify latest CDH results and can also be used as a benchmark for academic promotions or allocation of funding based on applied semi-qualitative research metrics. Over the past decades, CDH-related research has progressed from simple empirical observations to accumulation of best clinical evidence, becoming much more multidisciplinary with main research endeavors concentrating in a few high-income countries. Great strides in basic science and biomedical technology have contributed to a number of revolutionary new discoveries in the pathogenesis and pathophysiological mechanisms of CDH. Collaborative research has led to substantial progress in prenatal diagnostics and interventions, implementation of standardized neonatal treatment protocols, and most recently regenerative medicine therapy. All these advances hold now the promise of improving CDH patient care and outcome in the 21st century. International collaborations should, therefore, be strengthened to allow further evolution in this field.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent For this type of study, informed consent was not required.

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