

BIBLIOMETRIC ANALYSIS OF NEGLECTED TROPICAL DISEASE LITERATURE

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## **Abstract**

*Objective:* There is a disparity between burden of disease and research efforts. This is especially true in tropical medicine, particularly concerning neglected tropical diseases (NTDs). This paper reports on a bibliometric study of NTD research from the past five years, examining disparities between research on individual NTDs and between income groups. To aid in NTD research priority setting and to inform future NTD policy making, it is important to know how much research is being produced, the topics of research, and who is producing the research.

*Methods:* To assess current research trends and disparities in research, this study used Scopus SciVal, a bibliometric analysis tool powered by Elsevier, and Web of Science's bibliometric analysis tool, examining the scholarly output for the overall body of NTD research.

*Results and Discussion:* The data show that high mortality NTDs are the most researched, though incidence rates are lower than other NTDs, and high morbidity NTDs are still somewhat under researched. The data show that the NTDs with the highest degrees of international collaboration are decreasing in prevalence and burden. The data also show that high income countries produce the most research on NTDs, followed by upper-middle income countries. Low-income countries produce the body of work with the most international collaboration, followed by high income countries, indicating that these two income groups collaborate most frequently. Upper-middle income countries produce the research with the lowest international collaboration.

*Conclusion:* The main finding of this study is something quite intuitive and something the WHO and NGOs already know: international collaboration is the best way to combat NTD burden across the globe.

## Background

To neglect something is to fail to care for it, or to leave undone or unattended to [1]. In history, many areas of research could claim neglect as their own, as it has been well established that current research efforts are not meeting the world's health needs in a balanced way. More resources are allocated to health problems affecting high income countries, ignoring those diseases with higher burdens in lower income countries. The idea of the 90/10 gap in health research is still discussed, though the accuracy of the concept has been questioned in recent years, the general idea remains: only ten percent of global funding is being assigned to conditions affecting 90 percent of the world population [2]. This is true of many research areas within medicine, though as of yet the extent to which communicable infectious diseases have been researched is a relative unknown. There is enough research, however, to define a group of diseases as particularly underrepresented in scientific literature.

The World Health Organization (WHO) portfolio of neglected tropical diseases (NTDs) comprises a diverse group of 20 infectious diseases that predominantly affect the global south and people living under the poverty line. NTDs comprise of helminth infections, bacterial infections, viral infections, and protozoan infections, and snakebite envenomation, which is a slight outlier. Though these diseases are all epidemiologically and otherwise unique, they share the characteristic that they affect the most vulnerable among us. The burden of these diseases is exacerbated by poor living conditions and lack of infrastructure, which leads to unsafe waste management and lack of access to clean drinking water. NTDs perpetuate cycles of poverty through their high morbidity, mortality, and the deeply stigmatizing disfigurement resulting from some of these diseases.

In 2012, the WHO published an action plan to “accelerate work to overcome the global impact of neglected tropical diseases” [4] This work was the result of increasing coverage of NTDs by organizations such as Médecins Sans Frontières, an NGO focused on delivering healthcare to places and populations with vastly limited healthcare infrastructure. The plan had an initial portfolio of 17 diseases, and was expanded to include three more (taeniasis, scabies, and snakebite envenomation) by the 2021 action plan.

The 2012 action plan played a large role in kickstarting a global collaboration to combat NTDs, resulting in significant progress [5]. In 2022, half a billion people no longer require interventions for at least one NTD [6], and 46 countries have completely eliminated at least one NTD [7]. Measures to combat disease include increased research efforts as well as extensive collaboration with pharmaceutical companies to implement mass drug administration (MDA) programs. These programs are designed to empirically treat at-risk populations for a specific NTD caused by parasitic infection (e.g. taeniasis, soil-transmitted helminthiasis, schistosomiasis) or bacterial infection (e.g. Blinding trachoma, Buruli ulcer) within a defined geographic area. MDA programs are now the gold standard for NTD control in low- and middle-income countries with limited healthcare infrastructure [6].

In 2020, a new action plan was published, including bilharzia (schistosomiasis), Buruli ulcer, blinding trachoma, Chagas' disease, Dengue fever (and chikungunya), echinococcus, elephantiasis (lymphatic filariasis), foodborne trematode infections, Dracunculiasis (guinea worm disease), soil-transmitted helminthiasis (hookworm infections, ascariasis, tricuriasis), cutaneous and visceral leishmaniasis (kala azar), leprosy, mycetoma, rabies, river blindness (onchocerciasis), scabies, sleeping sickness (human African trypanosomiasis), snakebite envenoming, taeniasis (and cysticercosis), and yaws disease (Table 1). The CDC includes these same diseases in their action plans [8].

While the progress made over the last decade is notable, we know that the burden of NTDs is borne by over 1.7 billion people worldwide, and that almost all countries are endemic for at least one NTD [9]. These are likely under representations of the global NTD burden, as we do not have completely accurate estimates of disease burden and we do not have any literature mapping research efforts. At the moment, the WHO is mapping the delivery of treatments and new cases of NTDs, as well as producing estimates of disease burden in the form of mortality and disability-adjusted life years (DALYs) (Table 2).

Research efforts are not being consistently tracked in the field of NTDs, making it difficult to know whether current interventions are effective and are sufficient. A common concern is that, when an MDA program becomes implemented, the job is considered finished and the underlying causes of that NTD are left unaddressed, perpetuating the cycle of neglect [9].

To assess whether existing interventions are effective, research is necessary. To decide which research to fund, the current body of literature must be carefully assessed. Stakeholders (researchers, doctors, NGOs and IGOs, funding bodies, countries, etcetera) need to know where NTD research is being produced, which entities are collaborating to produce research, which diseases are being focused on more than others, and possible reasons for disparities between research efforts for different diseases and between different entities. As of right now, no such bibliometric research has been done. The WHO is keeping track of the chemotherapeutic interventions administered and who may still require these interventions [10].

## **Methods**

*Bibliometric methods for estimating research efforts and for bibliometric analysis.*

I relied on scientific publications to represent research efforts on NTDs from 2017-2022. These publications were collected from two places: Elsevier Scopus and Web of Science, using standardized MeSH terms that best describe the diseases in question, taking into consideration the multiple names for many NTDs, and filtering out undesirable results (see supporting documents). Despite the limitations of the data set, because Elsevier and Web of Science are limited in scope, I expect the data to reflect and capture overall research trends. I did not limit the data to only peer-reviewed articles, because oftentimes in NTD research and research priority

setting, case studies and other single-author publications reflect the reality of the situation in the absence of larger studies.

I used Web of Science and Scopus because both databases are equipped with analytic technology. I used Scopus SciVal to analyze scholarly output, to estimate international research collaboration and academic corporate collaboration, and then to look at citation count and scholarly output in the 10% citation percentile. I also was able to analyze the NTD scholarly output for different income stratifications. I used Web of Science for publication data, for example ranking publishers and institutions on their scholarly output, also looking at which publications are open access. I also used Web of Science for funding data, ranking the top funding agencies in the world. I used both to rank the top performing countries in the world. I then went through and analyzed the resulting datasets using the metrics below, comparing the data and rankings to disease burden estimates overall, based on geography, based on income level, and based on paediatric disease burden.

#### *Disease burden estimates*

To estimate the burden of disease, years of life lost (YYLs), I used years of life with disability (YLD), and disability-adjusted life years (DALYs) provided by the WHO in 2010 and 2015. I also used supplementary data provided by the WHO in subsequent years for diseases not classified as neglected at that time (Buruli ulcer, scabies, snakebite envenomation, guinea worm, mycetoma, yaws disease). These estimates were compared to the Though DALYs are not without their limitations, they are one of the most established methods of measuring disease burden outside of pure mortality figures.

**Table 1:** List of Neglected Tropical Diseases by Centers for Disease Control and Prevention (CDC) and World Health Organization (WHO).

<i>Disease</i>	<i>CDC</i>	<i>WHO</i>
<i>Buruli ulcer (Mycobacterium ulcerans infection)</i>	+	+
<i>Chikungunya</i> <sup>a</sup>	–	+
<i>Chagas disease</i>	+	+
<i>Cysticercosis</i>	+	+
<i>Dengue fever</i>	+	+
<i>Dracunculiasis (or guinea worm disease)</i> <sup>b</sup>	+	+
<i>Echinococcosis</i>	+	+
<i>Fascioliasis</i>	+	+
<i>Foodborne trematodes</i> <sup>a</sup>	–	+
<i>Human African trypanosomiasis (or sleeping sickness)</i>	+	+
<i>Leishmaniasis (or kala-azar)</i>	+	+
<i>Leprosy</i>	+	+
<i>Lymphatic filariasis</i> <sup>b</sup>	+	+
<i>Mycetoma</i>	+	+

<i>Onchocerciasis (or river blindness)</i> <sup>b</sup>	+	+
<i>Rabies</i>	+	+
<i>Schistosomiasis</i> <sup>b</sup>	+	+
<i>Soil-transmitted helminthiasis</i> <sup>b</sup>	+	+
<i>Trachoma</i> <sup>b</sup>	+	+
<i>Yaws</i>	+	+

Data source: CDC, 2017 [8]; WHO, 2017 [3]; <sup>a</sup> Not mentioned under CDC list of neglected tropical diseases (NTDs); <sup>b</sup> Diseases that can be controlled or eliminated through mass drug administration (MDA), or other interventions [11].

**Table 2:** Global Burden of Major Neglected Tropical Diseases as Estimates of Disability-Adjusted Life Years (DALYs), Years of Life Lost (YLL), and Years Lost Due to Disability (YLD) in WHO Member States [11].

Disease	2015 Data <sup>a</sup>		2010 Data <sup>b</sup>	
	YLL (thousand)	YLD (thousand)	DALY = YLL + YDL (thousand)	DALY (thousand) <sup>b</sup>
Soil-transmitted helminthiasis	449.50	3993.97	4443.47	5043
Ascariasis	225.30	869.37	1094.67	1254
Trichuriasis <sup>c</sup>	-	542.80	542.80	630
Hookworm <sup>c</sup>	-	1739.58	1739.58	3159
Schistosomiasis	1042.20	2471.65	3513.85	3971
Dengue fever	1848.79	761.29	2610.08	1243
Lymphatic filariasis <sup>c</sup>	-	2070.85	2070.85	2740
Cysticercosis	1258.27	598.09	1856.36	503
Rabies	1672.03	0.14	1672.17	2297
Leishmaniasis	1310.74	45.72	1356.46	3754
Onchocerciasis <sup>c</sup>	-	1135.57	1135.57	564
Foodborne trematodiasis	224.12	842.22	1066.34	665
Echinococcosis	568.20	73.23	641.43	600
Leprosy	457.67	30.97	488.64	215
Human African trypanosomiasis	368.68	2.97	371.65	1346
Trachoma <sup>c</sup>	-	278.97	278.97	308
Chagas disease	189.65	63.05	252.70	499

<sup>a</sup> Data source: WHO Health Statistics and Information Systems, 2015 [12]. Data on DALY are not available for Buruli ulcer disease; <sup>b</sup> Bhutta et al. (2014); <sup>c</sup> No data available for YLL.

\*Buruli ulcer, mycetoma, snakebite envenoming, and yaws are not included as there is not sufficient data from which to calculate DALYs.

**Table 3:** Number of Deaths and Age-Standardized Death Rates for NTDs in 2013 with Percent Change from 1990 to 2013.

NTDs	Deaths in 2013	Percent change since 1990	Age-standardized deaths per 100,000 in 2013	Percent change since 1990
Visceral leishmaniasis	62,500	19.8%	0.86	-0.3%
Rabies	23,500	-38.3%	0.34	-54.0%
Chagas disease	10,600	-19.3%	0.17	-51.7%
Dengue	9,100	-1.3%	0.13	-13.6%
African trypanosomiasis	6,900	-69.7%	0.08	-78.9%
Schistosomiasis	5,500	-68.2%	0.08	-80.7%
Ascariasis	4,500	-50.7%	0.06	-54.7%
Cystic echinococcosis	2,200	-45.0%	0.03	-60.8%
Cysticercosis	700	-28.6%	0.01	-53.0%
Hookworm*	0	NA	0	NA
Trichuriasis*	0	NA	0	NA
Foodborne trematodiasis*	0	NA	0	NA
Lymphatic filariasis*	0	NA	0	NA
Onchocerciasis*	0	NA	0	NA
Cutaneous/mucocutaneous leishmaniasis*	0	NA	0	NA
Trachoma*	0	NA	0	NA
Leprosy*	0	NA	0	NA
Other NTDs	16,300	-54.4%	0.24	-62.3%
<b>Total deaths from NTDs</b>	<b>141,800</b>	<b>NA</b>	<b>NA</b>	<b>NA</b>
Additional neglected diseases	Deaths in 2013	Percent change since 1990	Age-standardized deaths per 100,000 in 2013	Percent change since 1990
Typhoid fever	160,700	-10.8%	2.21	-25.9%
Venomous animal contact	57,200	-25.0%	0.82	-51.8%
Cholera	69,900	-44.3%	0.97	-51.9%
Paratyphoid fever	54,300	-14.9%	0.75	-28.0%
Cryptosporidiosis	41,900	-57.8%	0.58	-59.8%
Amoebiasis	11,300	-39.1%	0.18	-58.3%
Scabies*	0	NA	0	NA
Trichomoniasis*	0	NA	0	NA
<b>Total deaths from additional neglected diseases</b>	<b>395,300</b>	<b>NA</b>	<b>NA</b>	<b>NA</b>

\*Negligible evidence of deaths according to the Global Burden of Disease Study (GBD) 2013.

NOTE: For information on percent change calculations, see GBD 2013 capstone paper on mortality [4]. The estimates presented in this table are also available from the Institute for Health Metrics and Evaluation (IHME) website and previously published in [4]. **Abbreviations:** NA, non-applicable

<https://doi.org/10.1371/journal.pntd.0005424.t002>

### *Classification of countries by income*

I used World Bank data to classify countries as high income (HIC), upper-middle income (UMIC), lower-middle income (LMIC), and low income (LIC) [13]. I used the most recent 2022 data, because even though some countries have changed in classification over the past five years, the analysis of literature production by these countries is likely to reflect their change in status and average out over the past five years.

### *Classification of diseases by relative burden in different continents*

I relied on a wide range of data from the WHO, Global Health Observatory, the CDC, and independent research to classify diseases by their geography, summarized in Table 3. I also relied on various sources that compiled existing data. I marked the continents where a particular NTD is endemic with a positive sign.

**Table 4:** Global distribution of Neglected Tropical Diseases 2022.

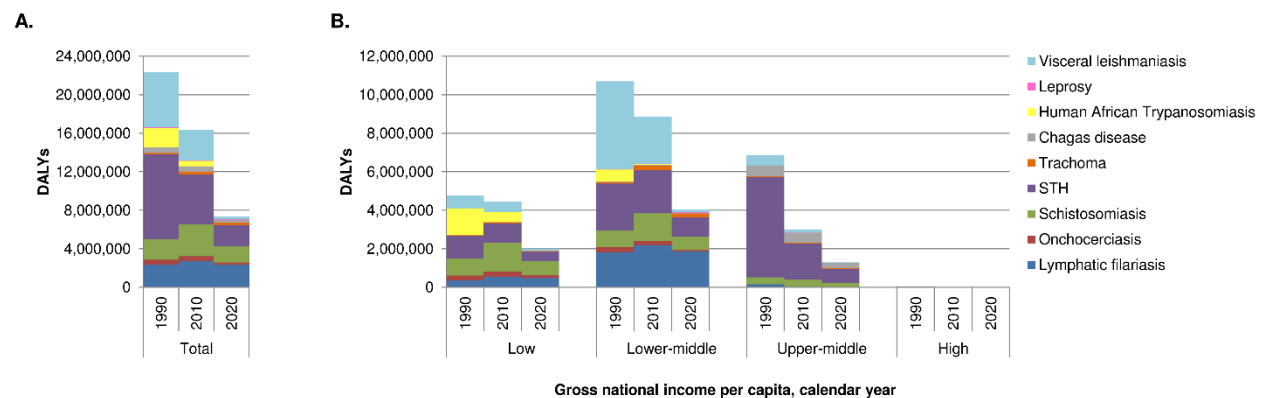
<i>Disease</i>	<i>Americas<sup>d</sup></i>	<i>Africa</i>	<i>Europe</i>	<i>Asia</i>	<i>Oceania</i>
<i>Soil-transmitted helminthiasis [14]</i>	+	+	–	+	+
<i>Schistosomiasis [15]</i>	+	+	–	+	–
<i>Dengue fever [16]</i>	+	+	–	+	+
<i>Lymphatic filariasis [17]</i>	–	+	–	+	–
<i>Taeniasis/Cysticercosis [18]</i>	+	+	+	+	+
<i>Rabies [19]</i>	+	+	+	+	–
<i>Leishmaniasis [20]</i>	+	+	–	+	–
<i>Onchocerciasis [21]</i>	–	+	–	–	–
<i>Foodborne trematodiasis [22]</i>	+	–	–	+	–
<i>Echinococcosis [23]</i>	+	+	+	+	–
<i>Leprosy [24]</i>	+	+	+	–	–
<i>Human African trypanosomiasis [25]</i>	–	+	–	–	–
<i>Trachoma [26]</i>	+	+	–	+	–
<i>Chagas disease [27]<sup>e</sup></i>	+	–	–	–	–
<i>Scabies [28]</i>	–	–	–	+	+
<i>Guinea worm [29]</i>	–	+	–	–	–
<i>Mycetoma [30]</i>	+	+	–	+	–
<i>Taeniasis/cysticercosis [31]</i>	+	+	+	+	+
<i>Snakebite envenomation [32]</i>	+	+	–	+	+
<i>Yaws [33]</i>	+	+	+	+	+

<sup>d</sup>Americas included as one entity because most NTDs in America will affect South and Central America, and sometimes stretch into the southern part of the United States of America.

<sup>e</sup> Because of the unique epidemiology of Chagas’ disease, people can become infected and then emigrate from their home country (most often endemic countries in Latin America) and continue to live with the disease in another country, which complicated the categorization. I elected to include only the Americas because that is where the disease most often will have originated. [27]

I also used existing research on between-country discrepancies in NTD burden.

**Fig. 1:** DALYs of top NTDs as compared to income level [34].

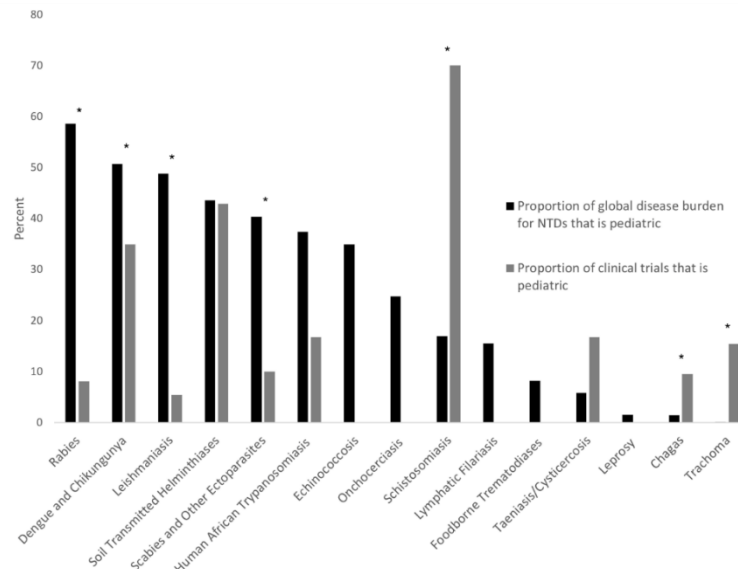


Stolk WA, Kulik MC, le Rutte EA, Jacobson J, Richardus JH, et al. (2016) Between-Country Inequalities in the Neglected Tropical Disease Burden in 1990 and 2010, with Projections for 2020. PLOS Neglected Tropical Diseases 10(5): e0004560. <https://doi.org/10.1371/journal.pntd.0004560>

## Paediatric Burden

I classified the diseases by their paediatric burden. Beyond this, categorization becomes more difficult. The individual could become infected as a child and then be faced with a lifetime of disability, making it difficult to classify it as a disease of childhood, adulthood, or old age.

**Fig. 2:** Paediatric global disease burden compared to the proportion of clinical trials that are paediatric [35].



Rees, Chris A et al. "Neglected tropical diseases in children: An assessment of gaps in research prioritization." *PLoS neglected tropical diseases* vol. 13,1 e0007111. 29 Jan. 2019, doi:10.1371/journal.pntd.0007111

## Research Collaboration

There is an established correlation between collaboration on research and citations of that research, particularly international collaboration [36]. To assess the extent to which this is the case in NTD research, and to assess which entities are most influential in deciding what to research, I have benchmarked the international collaboration of NTD research in Scopus. Another important factor in global health and medicine is the corporate machine, most notably pharmaceutical companies. I was able to benchmark academic-corporate collaboration for NTD research in order to assess the extent to which the corporate world influences NTD research prioritization.

## Funding Metrics: Top Funding Agencies, Grant Numbers

I used Web of Science to analyze the body of research produced using the standardized MeSH terms. I looked for top publishing agencies, and top performing grant numbers. The grant numbers posed a bit of a challenge because they were not listed along with the organization providing the grant. I was able to look at the individual publications in each of the top grant sections, where the grant number and funding agency were listed.

## Results

*Scopus SciVal: Scholarly Output, International and Corporate Collaboration, and Citation Count.*

**Fig. 3:** Scholarly Output vs. Publication Year of All Neglected Tropical Diseases from 2017-2022

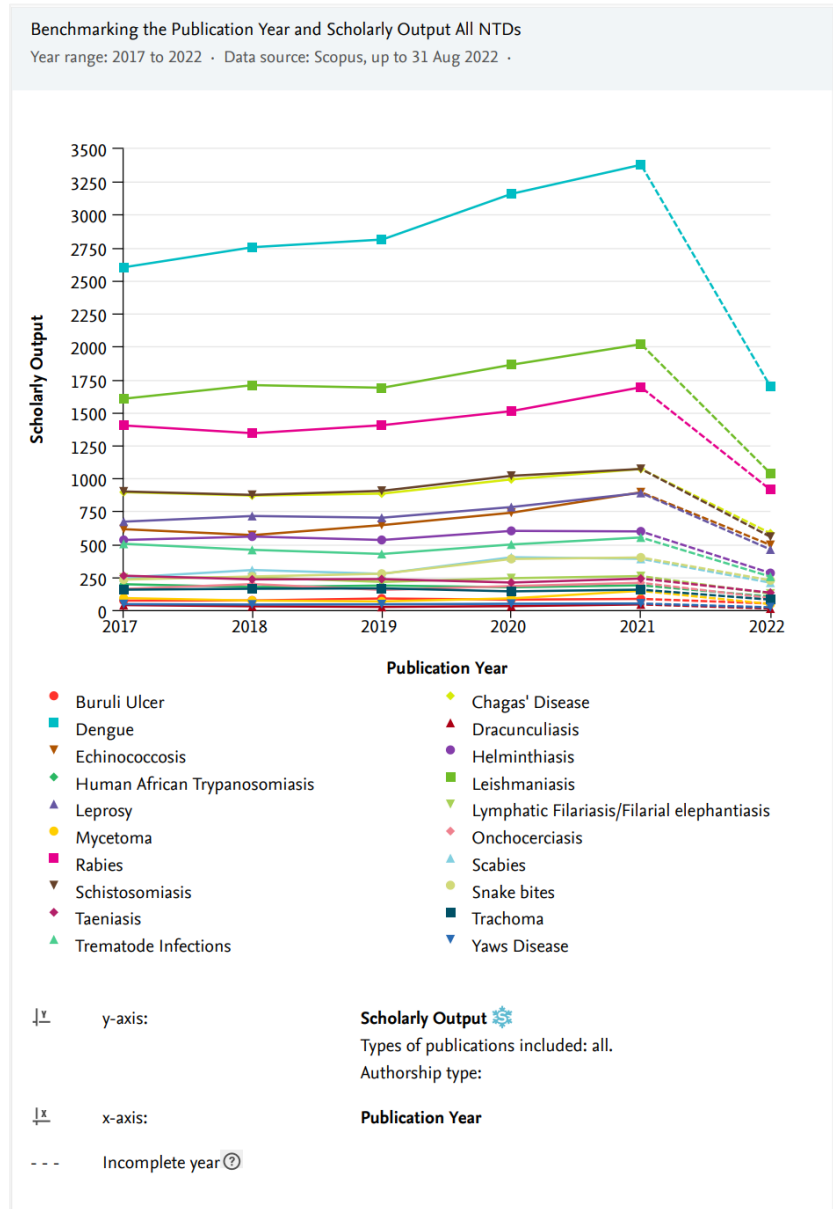


Figure 3 indicates that Dengue fever is the most researched NTD by a significant margin. It is followed by Leishmaniasis and Rabies. Dracunculiasis (guinea worm disease) is the NTD with the lowest scholarly output over the past five years, followed by Yaws disease and Buruli Ulcer. We can also see an overall increase in scholarly output for all NTDs over the past five years, which is promising. The rate that scholarly output is increasing is relatively consistent between all of the NTDs.

**Fig. 4:** Scholarly Output vs. Publication Year of NTD Research Produced by Different Income Groups from 2017-2022

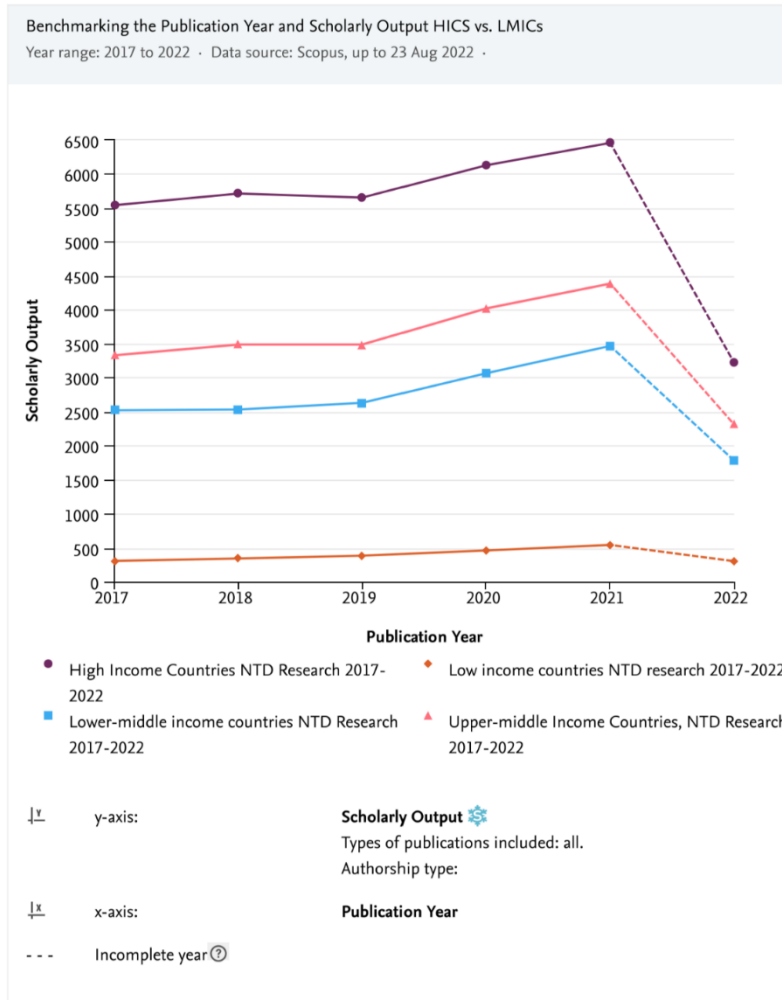


Figure 4 depicts the scholarly output of NTD research of high income, upper-middle income, lower-middle income, and low-income countries from 2017-2022, so the past five years. Predictably, high income countries make the largest contribution to the body of literature, and the number of publications decreases with income. Middle income countries still contribute quite significantly to the body of knowledge, and the research published by low-income countries is still statistically significant.

**Fig. 5:** International Collaboration (%) vs. Publication Year of NTDs.

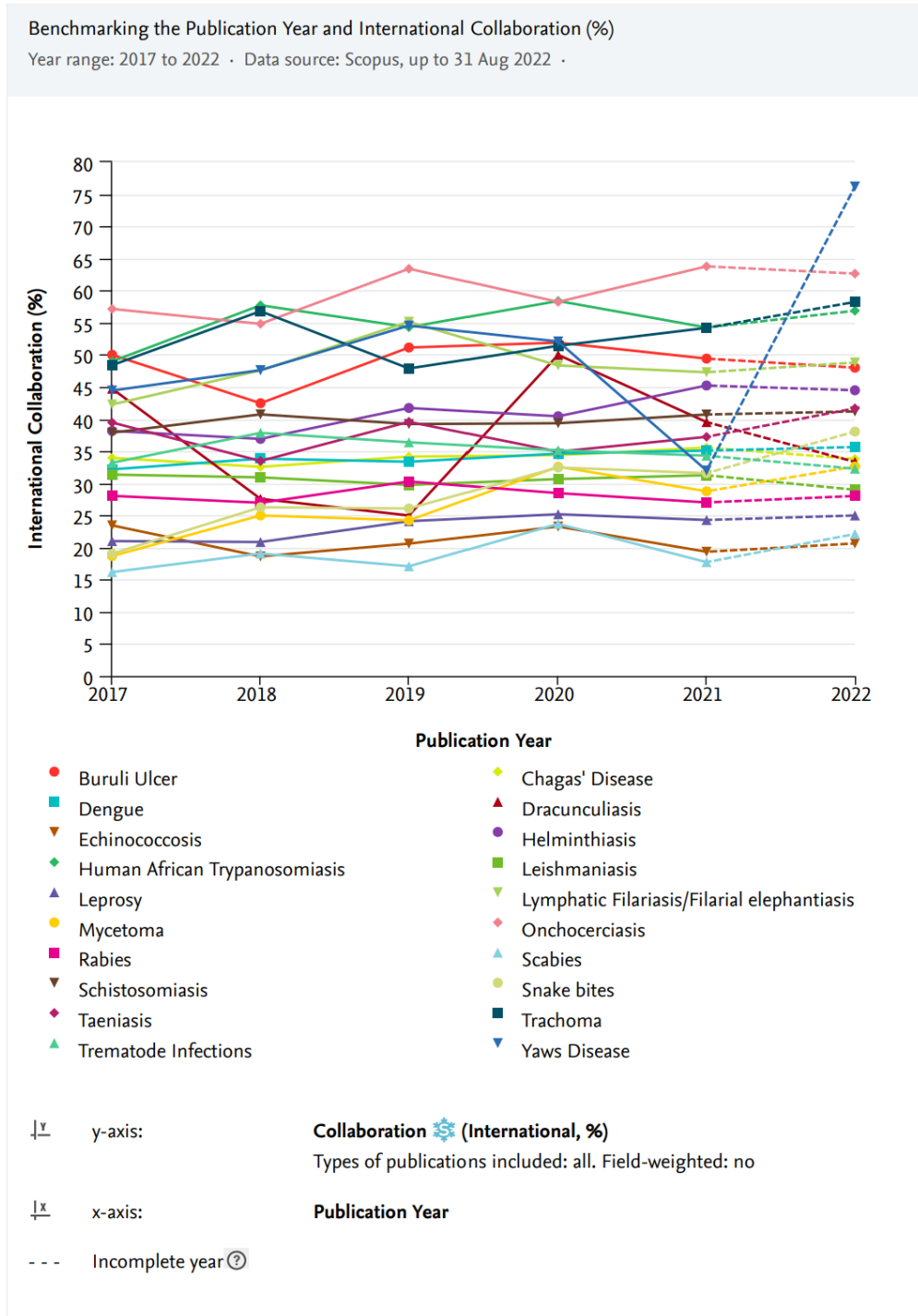
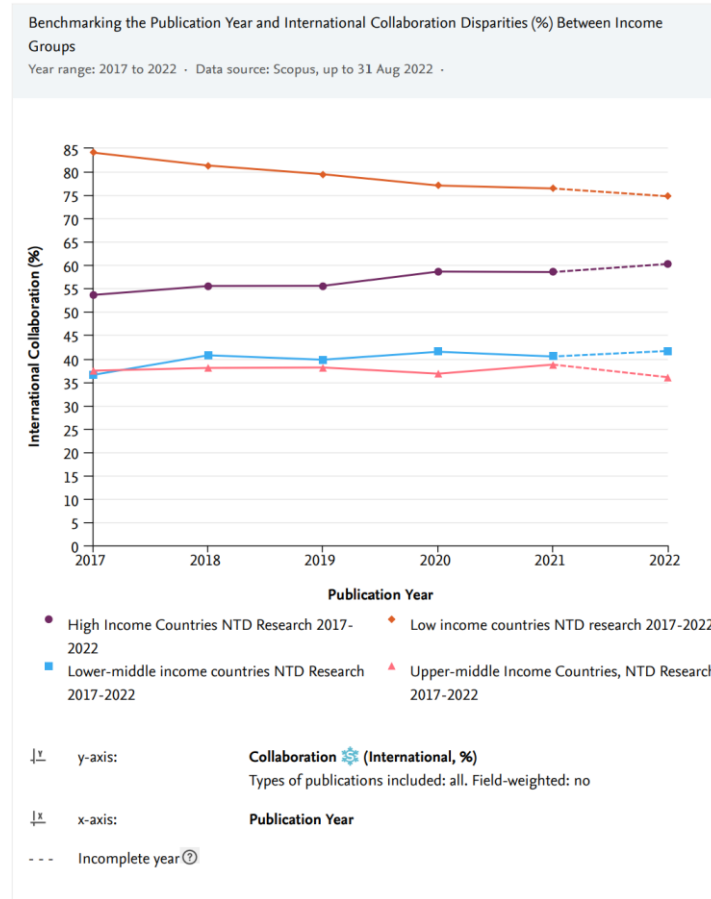


Fig. 5 is a bit messier and more difficult to interpret; however, it still contains valuable insights. Onchocerciasis (river blindness) has the greatest overall international collaboration, followed by Human African Trypanosomiasis (sleeping sickness), and then Trachoma.

**Fig. 6:** International Collaboration of NTD Research Produced by Different Income Groups (%) vs. Publication Year from 2017 to 2022



**Table 6:** Overall International Collaboration of NTD Research Produced by Different Income Groups (%) vs. Publication Year 2017 to 2022

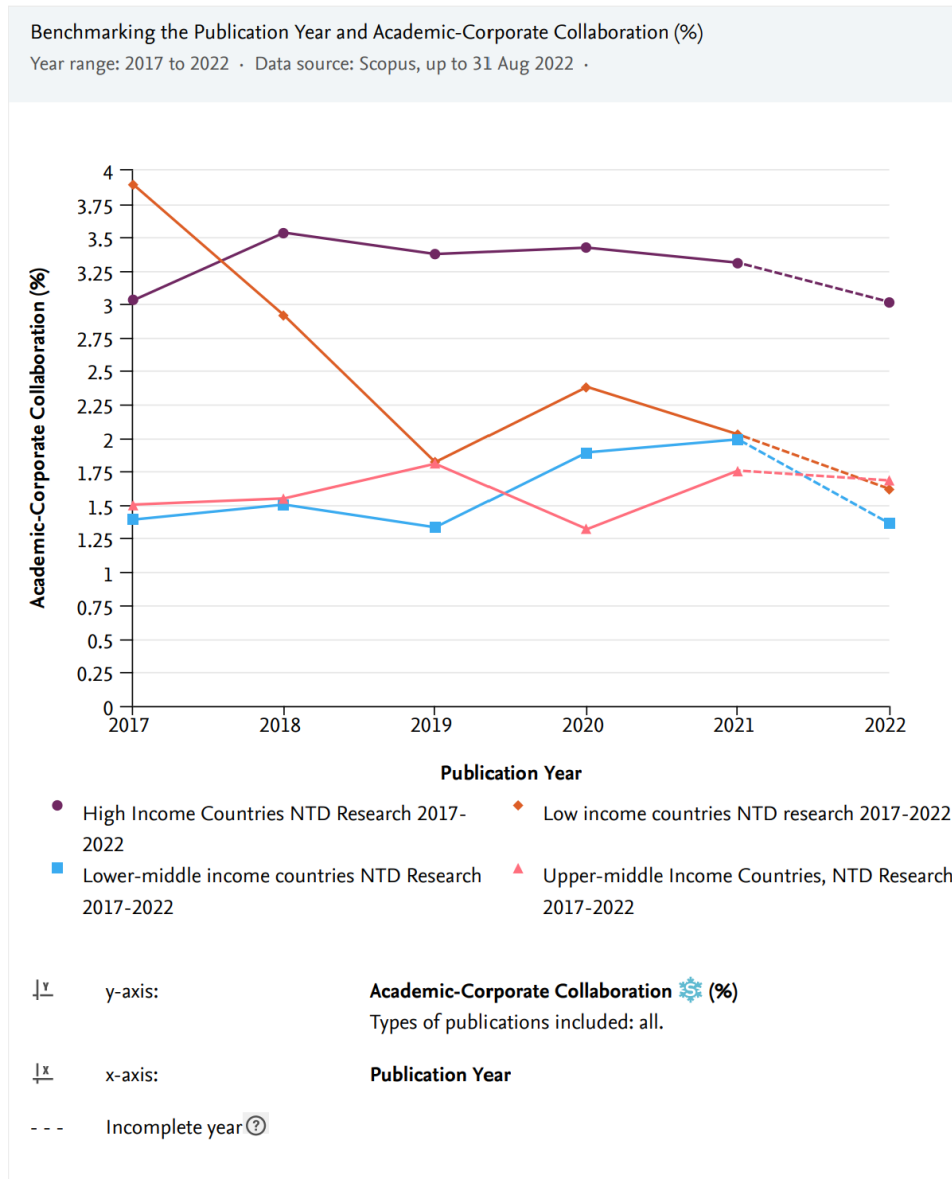
Benchmarking the Publication Year and International Collaboration (%)  
Year range: 2017 to 2022 · Data source: Scopus, up to 31 Aug 2022 ·

Entity	Overall ↓
Low income countries NTD research 2017-2022	78.6
High Income Countries NTD Research 2017-2022	56.9
Lower-middle income countries NTD Research 2017-2022	40.1
Upper-middle Income Countries, NTD Research 2017-2022	37.6

Metric 1: Collaboration (International, %)  
 Types of publications included: all. Field-weighted: no  
 Metric 2: Publication Year

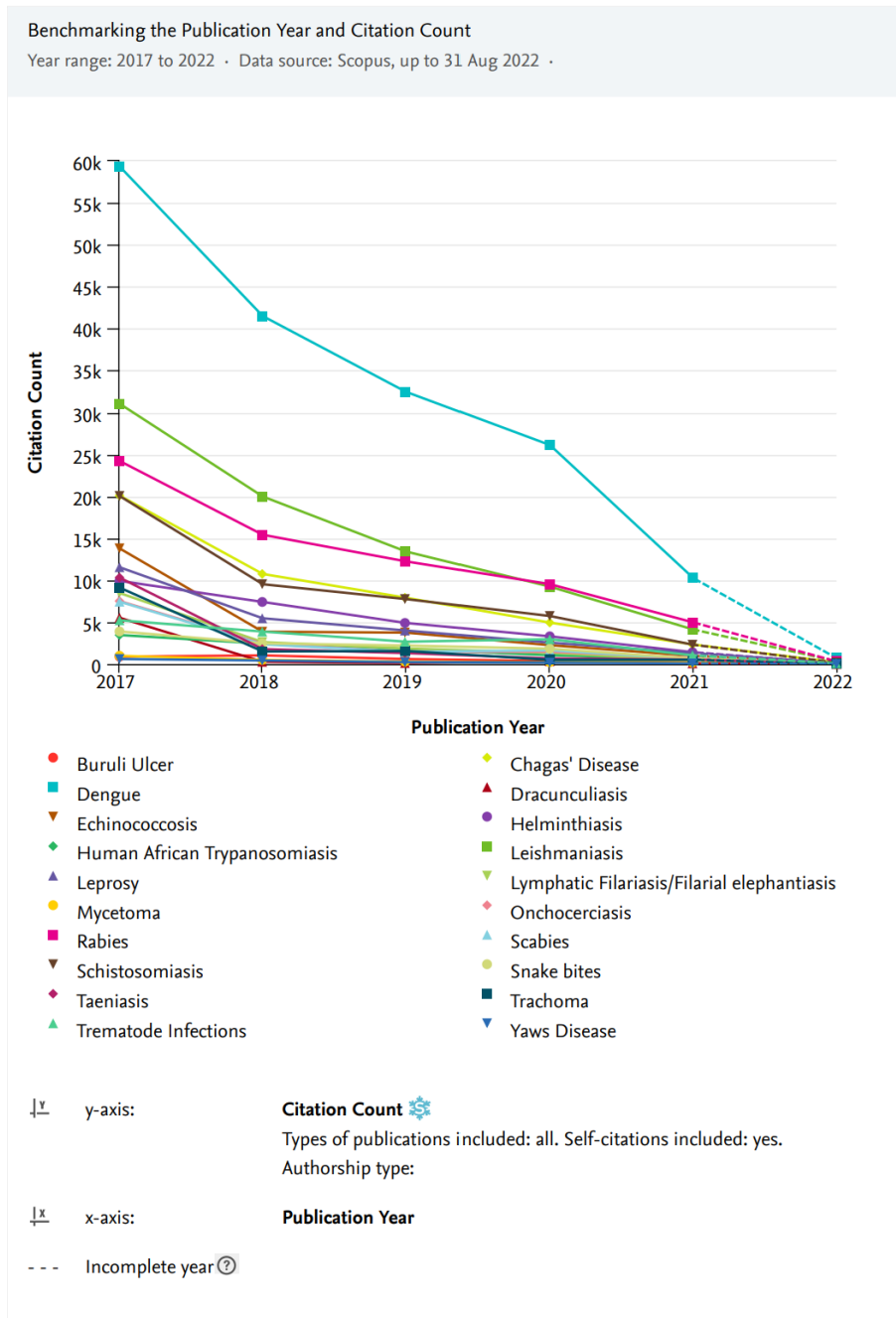
Fig. 6 depicts the percentage of publications from the overall scholarly output from each income group that is the product of international collaboration. Low-income countries have the highest degree of international collaboration with an average of 78.6% of publications being a result of international collaboration. High income countries follow with 59.6% of publications, then lower-middle income countries with 40.1%, and then upper-middle income countries interestingly are the lowest on this metric with 37.6% of publications (see Table 6). The extent to which countries are collaborating to produce NTD research is not changing much over time.

**Fig. 7:** Academic-Corporate Collaboration (%) vs. Publication Year HICs. Vs. LMICs from 2017 to 2022



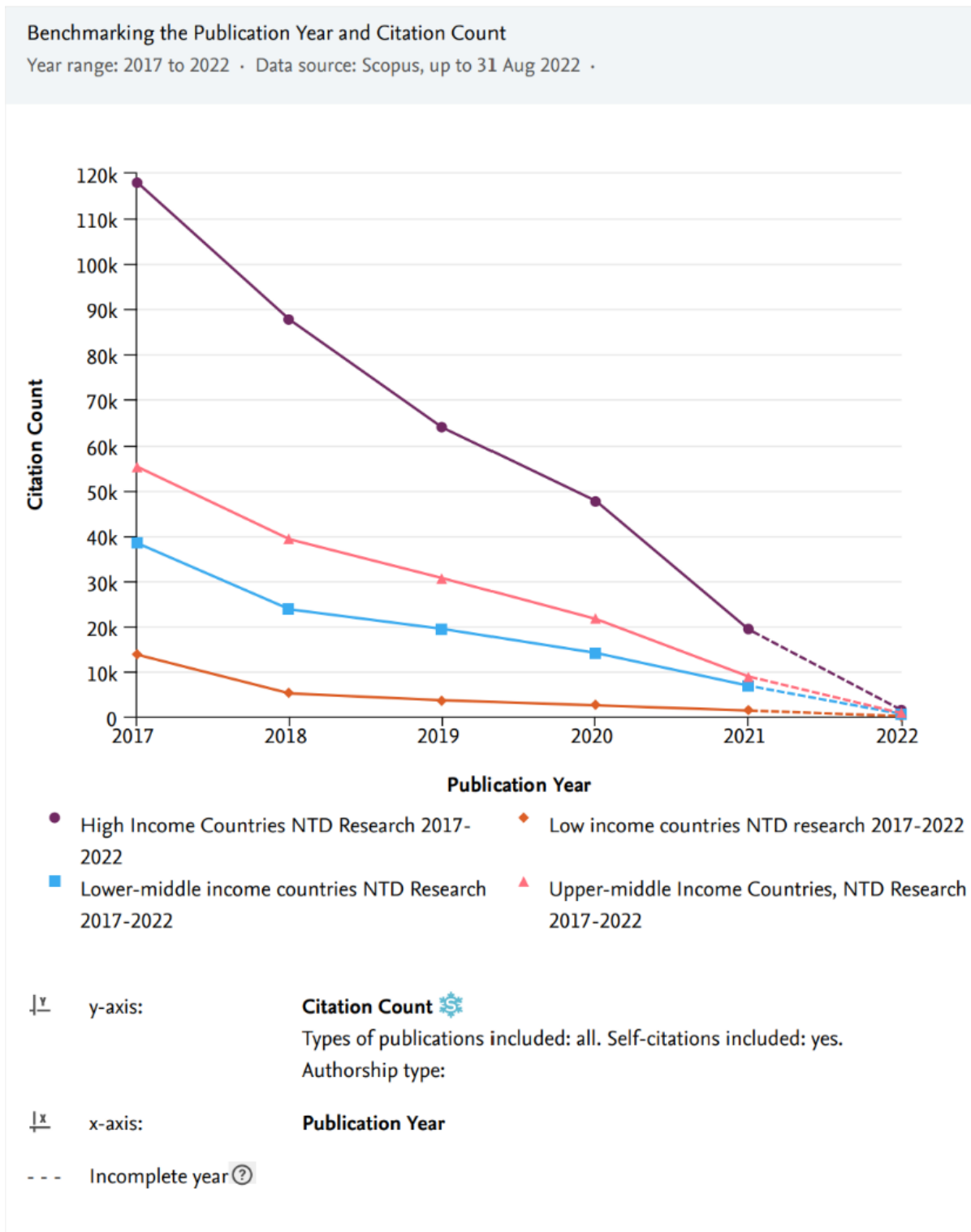
Predictably, high-income countries have the highest degree of academic-corporate collaboration, followed more interestingly by low-income countries. Considering the extent to which these two income groups collaborate to produce NTD research this is less surprising.

**Fig. 8:** Citation Count vs. Publication Year of NTD Publications from 2017 to 2022



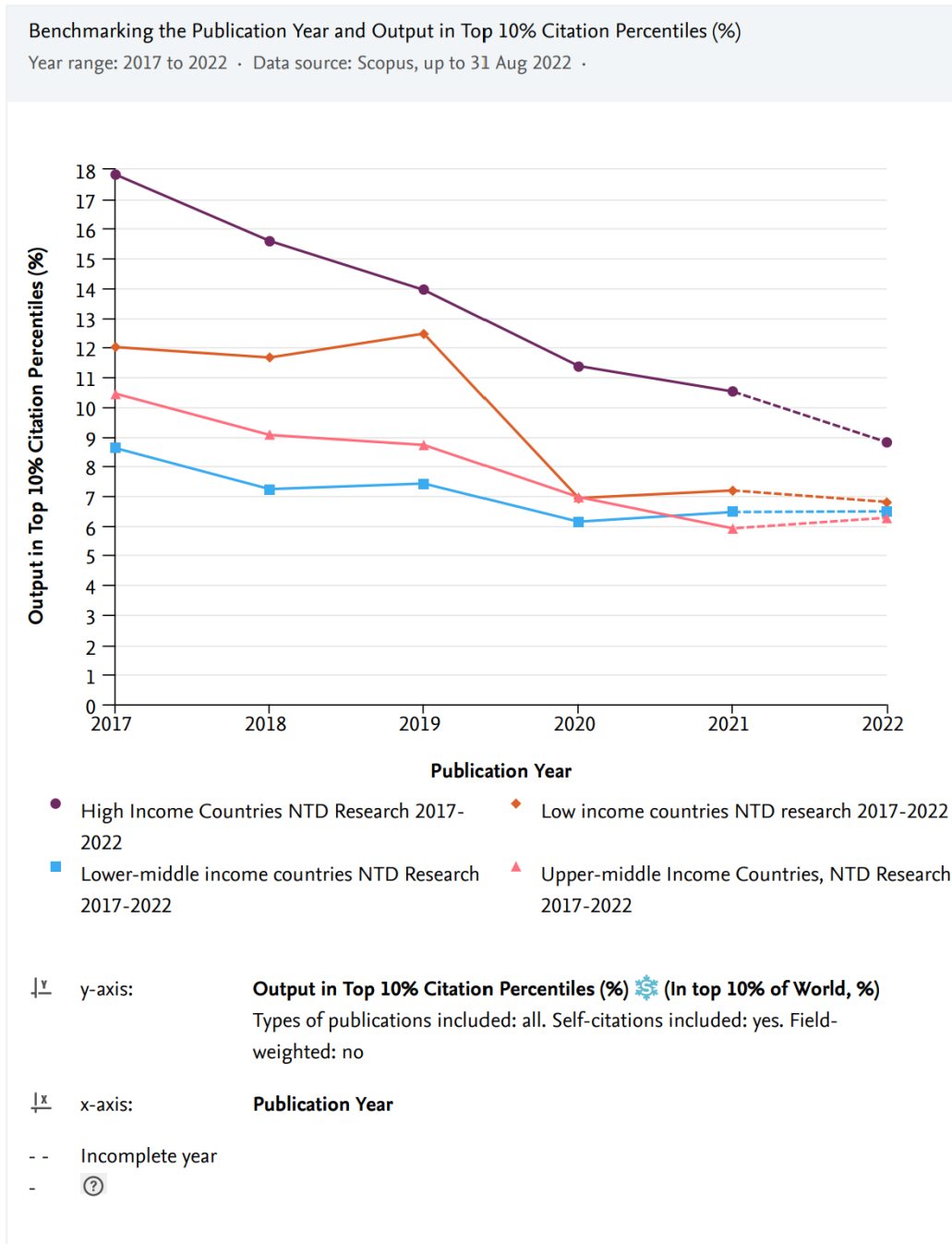
Overall citation counts of NTD research is decreasing, which is interesting because overall research is increasing. This could be because the field is growing and more new research is being produced, so the number of citations for papers on individual diseases is decreasing. This is still somewhat concerning, however, as it could indicate that NTD research is becoming less accessible, and less published in open-access journals. Dengue fever has the highest citation count, and Yaws Disease has the lowest.

**Fig. 9:** Citation Count vs. Publication Year of NTD Research Produced by Different Income Groups 2017 to 2022



Predictably, research produced by high income countries is cited the most, and the number citations decreases by income group. This follows the trend that high income countries produce the most research on NTDs, so they will have the highest number of citations.

**Fig. 10:** Output of NTD Research in the Top 10% Citation Percentiles vs. Publication Year Produced by Different Income Groups from 2017-2020.



This is perhaps a more accurate measure of the impact of the research produced by each income group. This depicts the percentage of research produced by each income group that ends up in the top 10% of publications, measured by their citation count. As previously stated, international collaboration and citation count have been established as having a positive correlation. This chart resembles the chart on international collaboration, indicating that this holds true for NTD research. The income groups with higher levels of international collaboration have a higher output of publications in the top 10% of citation percentiles.

*Web of Science: Top Funding Agencies, Top Producing Countries, and Top Publishers.*

Based on the standardized MeSH terms describing current NTDs, 14 732 publications were identified by *Web of Science* from the past five years (2017-2022).

**Fig. 11:** Scholarly Output of NTD Research vs. Country of Origin 2017-2022.

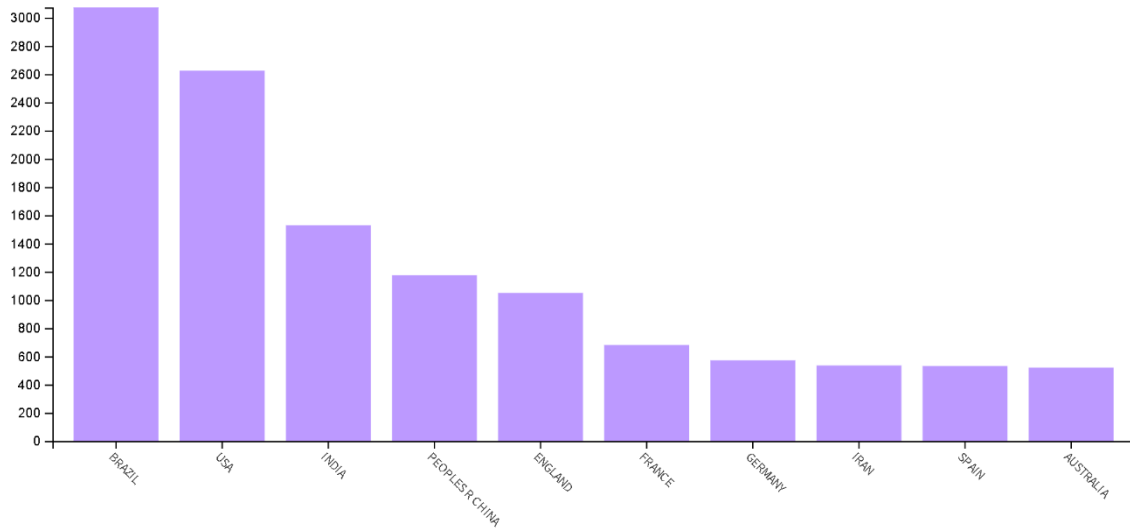


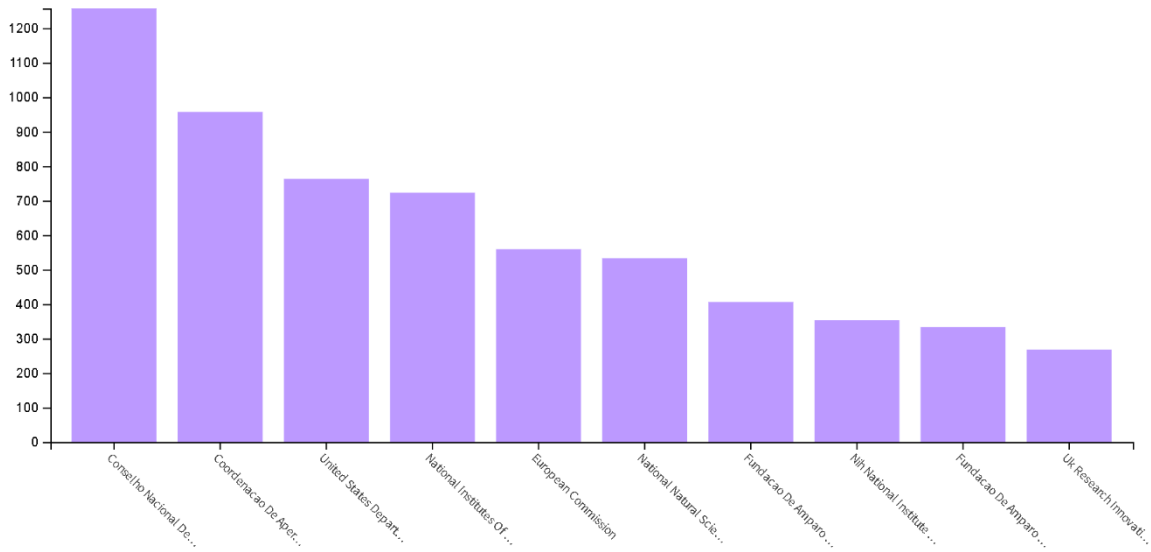
Fig. 11 shows the top performing countries in terms of scholarly output. Some of these results are interesting because, although HICs overall produce the most research on NTDs, some upper middle-income countries rank very highly, particularly Brazil. This is interesting because the research capacity of these countries is often underestimated by researchers and scholars in HICs.

**Fig. 12a:** Top Funding Agencies in the World TreeMap Chart 2017 to 2022.



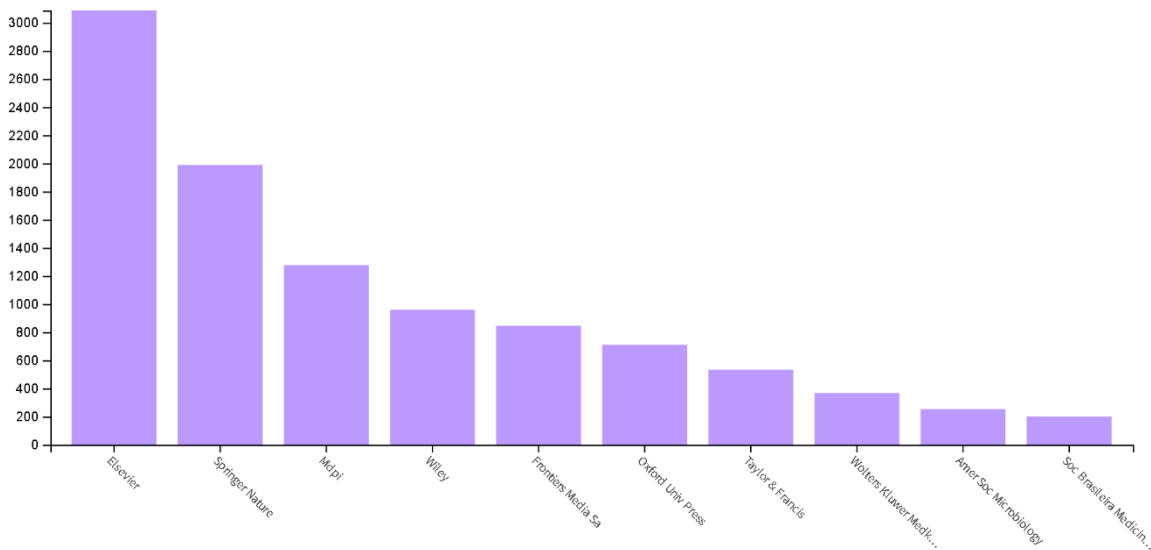
\*TreeMap Chart included because the bar chart does not provide complete names of the funding agencies.

**Fig. 12b:** Top Funding Agencies in the World Bar Chart 2017 to 2022.



From this, we can see several familiar names; however, the top two funding agencies are not from a high-income country but rather Brazil, which is an upper-middle income country. This is interesting considering that HICs produce the most research on NTDs.

**Fig. 13:** Scholarly Output vs. Publishers of NTD Research; Ranking Top Publishers 2017-2022.



This chart depicts the top publishers of NTD research. A lot of these are familiar titles, but some are less familiar and include non-English publications (e.g. *Soc Brasileira Medicina Tropical*). This is interesting because it indicates that English-speaking populations and researchers are not the only influential stakeholders when it comes to NTD research.

**Fig. 14:** How much NTD Research Published is Open Access 2017-2022.

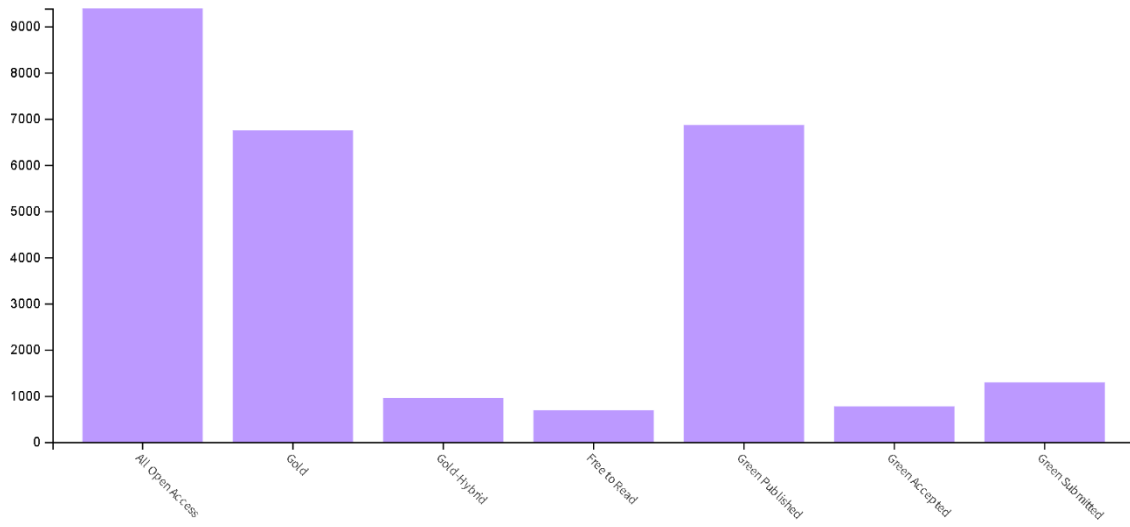


Fig. 14 shows how much of the literature published from 2017 to 2022 that is open access or stuck behind a paywall. A good proportion is open access (63.7%), but a lot of research is not open access. This is significant because the proportion of publications that are open access impacts the citations of NTD research.

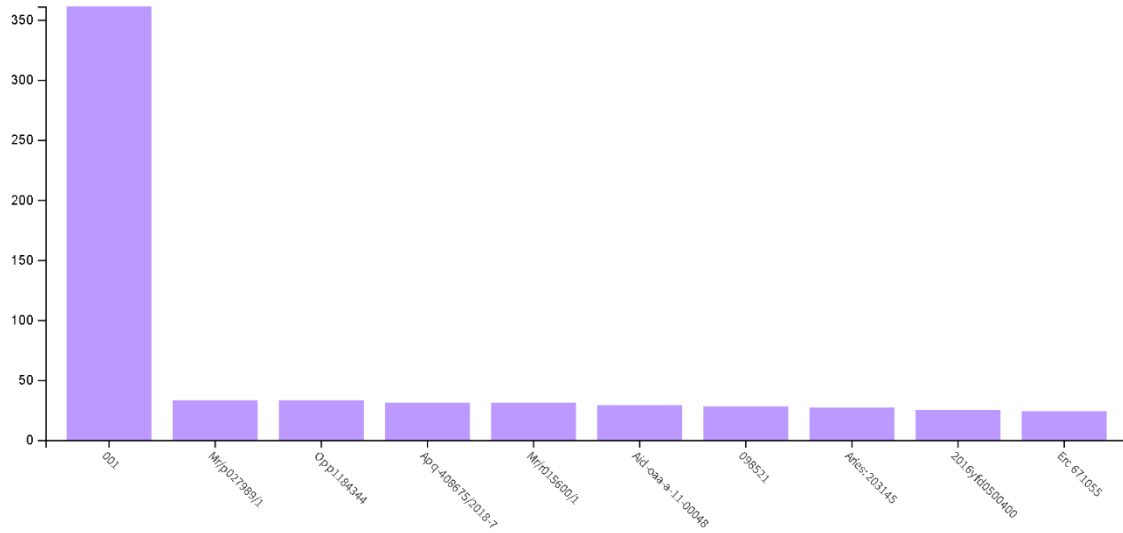
**Fig. 15:** Agencies Funding Open-Access Publications from 2017 to 2022.



Fig. 15 analyzed the open access column from Fig. 14, so the body of research that is open access, and shows the top funding agencies of open-access publications. This is significant

when compared to the previous funding table, as we can see that American agencies rank higher than some Brazilian agencies, and two of the top three funding agencies are in fact American.

**Fig. 16:** Number of Grants awarded to NTD research from 2017 to 2022



\*this is measured using grant numbers. Each grant awarded has a specific number associated with it, which is listed along with the funding agency in the Web of Science database.

This metric is not the most useful because the name of the grant and funding agency are not published along with the grant number. I was able to find the agency funding the grants, however. The most important is 001, which is a WHO grant. The others are less significant and align with previous high performing funding agencies.

## Discussion

In this document, I have analyzed the existing body of NTD research, breaking it down by disease type and income level. Using SciVal, I then ranked these sections of NTD literature based scholarly output (Fig. 3, Fig. 4), international and corporate collaboration (Fig. 5, Fig. 6, Fig. 7, Table 6) and citation count metrics (Fig. 8, Fig. 9, Fig. 10). Using Web of Science, I then looked at the top producing countries (Fig. 11), top funding agencies (Fig. 12a and 12b), top producing agencies (Fig. 13) and top publishing agents (Fig. 14). I looked at what proportion of NTD research is open access (Fig. 15), and what agencies are funding open access research (Fig. 16). Finally, I looked at grants awarded to NTD research, finding that the WHO awarded the most grants.

### *NTD Scholarly output over time: influenced by morbidity or mortality?*

When we look at Fig. 3, all of the NTDs ranked in terms of scholarly output, we see some interesting patterns emerge when compared against some of the morbidity and mortality data. As we can see from some WHO reports and from publications compiling WHO data on disease burden and mortality rates, diseases with high mortality have a greater scholarly output over time. Five of the six most deadly NTDs top the chart for scholarly output over time: visceral leishmaniasis, rabies, chagas' disease, dengue haemorrhagic fever, and schistosomiasis [19, 37]. Obviously, diseases that have a high mortality and affect everyone are prioritized, for example Dengue and Rabies. Rabies has a relatively low baseline incidence in comparison to Dengue but has the potential 100% deadly if not treated extremely quickly [19]. Rabies has also had a fair bit of media coverage, which could also have affected the scholarly output along with citation rates [38].

Some of the top ranked diseases in terms of scholarly output, along with having high mortality rates, are quite dramatic in their clinical presentation. This could contribute to the high scholarly output on these diseases because cases are easy to identify and easy to study. These diseases look scary and are scary. Dengue haemorrhagic fever (the most severe form of Dengue) in particular can have very alarming symptoms, including bleeding from the nose and gums, and vomiting blood [39]. Rabies is also very dramatic. Symptoms of clinical rabies include delirium, hallucinations, fear of water, paralysis, and excessive salivation [40]. Visceral leishmaniasis (kala azar) has a mortality rate of 95% without treatment, and symptoms include persistent fever and extreme anorexia, resulting in emaciated look that is highly identifiable [41].

Diseases with high morbidity and high incidence rank just below diseases with high mortality on scholarly output. Foodborne trematode infections and helminth infections rank highly, despite having a low mortality rate. They do, however, have high DALYs (Fig. 1, Table 2), which likely contributes to their larger scholarly output. However, diseases that cause identifiable disfiguration along with high morbidity rank higher on scholarly output. Lymphatic filariasis and leprosy are perhaps the most disfiguring, and rank highly in terms of DALYs

(Table 2). Leprosy, of all of the non-fatal diseases, ranks the highest in terms of scholarly output in the past five years (Fig. 3).

### *Paediatric Disease Burden and Scholarly Output*

Rabies, Dengue, and leishmaniasis are the NTDs with the highest disease burden (Fig. 2). These are the NTDs with the highest mortality (Table 3), and the diseases that are ranked highest in terms of scholarly output (Fig. 3). Overall, the ranking of diseases by paediatric burden is similar to scholarly output. Helminthiasis also has a high paediatric burden and is ranked highly in terms of scholarly output. This is likely because NTDs disproportionately affect children in terms of DALYs and mortality [42]. It is promising that current research trends reflect this.

### *International Collaboration on NTD Research*

When looking at the percent of research for each NTD produced as a result of international collaboration (Fig. 5), we see that sleeping sickness ranks very highly, along with blinding trachoma and river blindness (onchocerciasis). All are examples of NTDs that have seen significant progress over the past decade and are WHO road map targets for elimination as public health threats by 2030 [6].

One disease that did not rank as highly as maybe expected despite considerable mortality is human African trypanosomiasis. Sleeping sickness is characterized largely by behavioural symptoms, because the disease-causing parasite eventually is able to cross the blood-brain barrier and infect the brain. Aggressiveness, decreased inhibition, poor coordination, and trouble sleeping at night but desire to sleep during the day are all characteristics of the late stage of disease. This disease is the fifth most deadly NTD (as of 2013) [25], yet ranks seventh from the bottom in terms of scholarly output from 2017 to 2022 (Fig. 3).

This is the result of interventions gone right, due to collaboration between the WHO, national control programs, and NGOs. Médecins Sans Frontières (MSF) in particular was instrumental in helping to screen for sleeping sickness, and for helping to organize healthcare delivery to affected areas [41]. From when MSF started screening for sleeping sickness in 1986 to 2019 when they ended the sleeping sickness team's activities, reported cases dropped from nearly 40,000 at a peak in 1998 to only 663 in 2019. Trachoma is another good example of international collaboration gone right. Since 2012, 12 countries have eliminated it entirely. Trachoma is the target of extensive MDA programs, with 32 million people receiving antibiotics in 2020 alone.

These are significant improvements indicating that international collaboration and cooperation between NGOs and IGOs along with national bodies is necessary to achieve meaningful change in the NTD sphere.

### *Scholarly Output and International Collaboration on NTD Research by Different Income Groups*

When I stratified countries based on their economies, according to the World Bank data (High income, upper middle income, lower middle income, and low income) I found that high income countries produce the most research on NTDs overall, followed by upper middle-income countries, then lower middle-income countries and low-income countries. This was expected.

When I looked at international collaboration for each of these income groups, low-income countries have the highest rate of international collaboration with 78.6% of overall NTD research being produced as a result of international collaboration (Fig. 6). This is likely because high income countries have a low NTD burden (Fig. 1), so to study NTDs, researchers from high income countries need to collaborate with low-income countries. Low-income countries do not have the research capacity to conduct research independently, so they turn to high-income countries.

Interestingly, upper middle-income countries have the lowest overall rate of international collaboration, with under 40% of research produced as a result of international collaboration. This is likely because these countries are affected by these diseases, so NTDs are prioritized in national funding. They have the money to invest in NTD research and they also have NTD patients in the country, so collaboration is not as necessary to produce quality research.

When we look at the countries with the highest scholarly output, Brazil is at the top (Fig. 11). This is interesting because it is an upper-middle income country, rather than a high-income country. High income countries often forget that upper-middle income groups have significant research capacity, and it is interesting to find a field in which a middle-income country is the leading producer of research.

Overall, diseases affecting the Americas have the most research done (Table 4, Fig. 3). When we consider that the countries with the highest scholarly output are the USA and Brazil, it becomes clear why this is the case.

### *Citation Count and Collaboration*

As has already been established, there is a correlation between international collaboration on research and citation count, which may incentivize researchers to collaborate to a greater extent than they otherwise would [36]. This is the case when we compare the extent to which different income groups collaborate on NTD research and scholarly output in the top 10% citation percentiles (%) (Fig. 10), rather than just raw citation count (Fig. 9). The ranking of income groups using this metric is the same as the ranking by international collaboration on NTD research (Fig. 6), and the ranking by academic-corporate collaboration (Fig. 7). However, less than four percent of research on NTDs is produced as a result of academic-corporate collaboration. Considering that the primary interventions for NTDs are MDA programs, it is concerning that corporations, the ones supplying the drugs for these programs, are not taking

more of an interest in NTD research to make sure the drugs they are donating and the programs they are contributing to will actually be effective in combatting NTDs.

This indicates that, predictably, high income countries have a great deal of decision-making power when it comes to NTD research; however, low-income countries seem to play a very influential role in the field despite have a much lower scholarly output, based on scholarly output in the top 10% citation percentile. This is good because traditionally non-HICs have been sidelined in academic discourse [43], and indicates a shift away from the 90/10 gap concept and towards an academic community that welcomes knowledge from diverse sources.

Citation count overall is dropping (Fig. 8, Fig. 9, Fig. 10); people are citing research on NTDs less, even as overall scholarly output is increasing (Fig. 3, Fig. 4). This is concerning, as it means the impact of this type of research could be dropping. However, there is a possibility that the field is changing. There is a movement away from discussing individual NTDs, and towards integrated treatment programs [6]. The metrics I used to measure citation count may not be picking up on the high-impact papers in the field. I was not able to find a single reason for this drop in citation count over the past five years. The COVID-19 pandemic likely played a role: there is a sharp drop in citation count in 2020.

#### *Funding Agencies and Grants*

Interestingly, the top funding agencies of NTD research are Brazilian, followed by American and European funding agencies (Fig. 12). This aligns with the top producing countries. This indicates that Brazil is investing heavily in NTD research and should be considered more as a leading authority in the field. When considered alongside the international collaboration metric, we can see that upper middle-income countries do not collaborate on research to the same extent as high- and low- income countries. It would be beneficial for high- and low- income countries to increase collaboration with middle income countries because these countries have considerable history in combatting NTDs, so they will likely have a greater understanding of what needs to be prioritized in NTD research.

The WHO is entity awarding the highest number of grants at the moment, which is not very surprising (Fig. 16). Considering that it is not in the top funding agencies, however, it likely gives a lot of small grants rather than a few smaller grants.

#### *Top Publishers of NTD Research and Open Access*

The top publishers of NTD research are from high-income countries (Fig. 13). Elsevier is a Netherlands based publication and Springer Nature is American. These are both high-impact journals. Frontiers ranks lower. These are all very high-impact journals with very good reputations [44]. A lot of NTD research is open-access, which is a good thing, but a significant proportion is still stuck behind a paywall (Fig. 14). Overall, the USA funds more open access publications (Fig. 15). This is another reason why increased collaboration with middle income

countries is necessary: if countries like Brazil collaborate with high- and low- income countries, then their research is more likely to be published in open-access, high impact journals.

### *Limitations and Further Research*

My findings likely do not capture the full extent of NTD research, or the burden of NTDs. Though NTD disease surveillance has improved in recent years, all estimates likely fall quite far short of the true numbers [6]. In addition, the full body of NTD research was likely not included in this study because many NTDs are referred to as many different names, both colloquially and in their literature. It is likely that some studies refer to diseases by different names and therefore did not show up.

Further research on citations would be useful to assess the drop in citations over the past five years. More work on citations and the field as a whole will be needed to examine the impact of existing publications and the impact new publications play. A literature review is likely needed to gain a greater understanding of the NTD field.

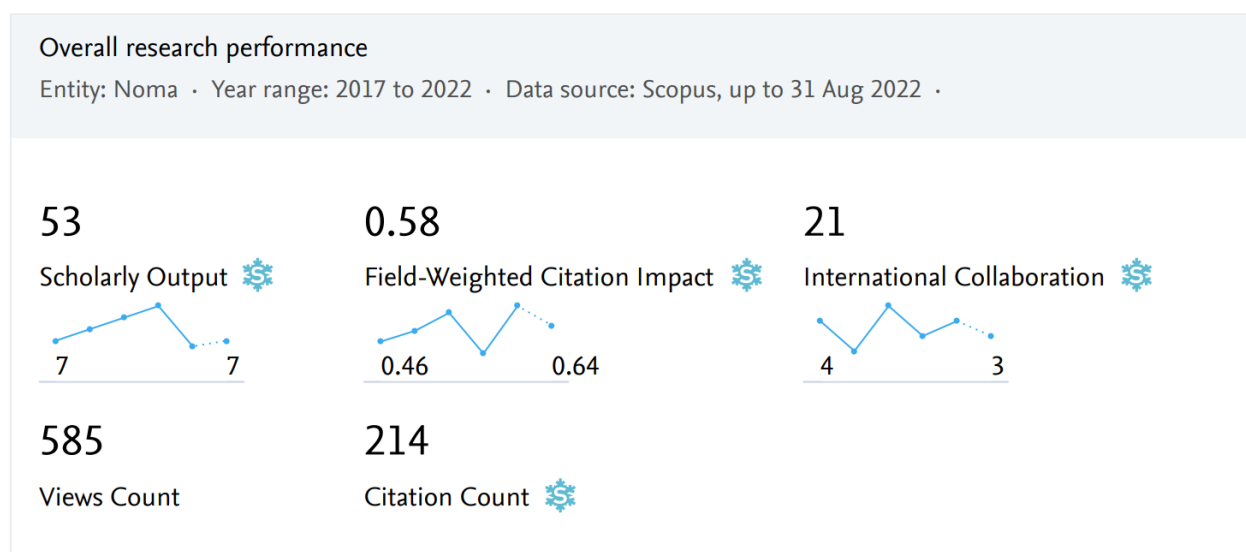
A more thorough examination of the literature is needed on how research prioritization and scholarly output relates to demographic, beyond paediatric disease burden because I was not able to assess this in this study.

I was not able comprehensively examine grant information or funding data of NTDs. I was very difficult to find this information, and what information I did find was limited in scope, only containing the number of grants an agency awarded rather than the amount and the time scale. More transparency is needed when it comes to research funding so that we can see what is being funded and by who.

### **Noma**

Noma is a little-known neglected disease that affects children living in poverty. It is found in low-income countries in Africa and Asia with limited access to healthcare. It is highly disfiguring, destroying the jaw, lips, cheeks, nose, or eyes. The most recent estimates on the global burden of Noma are more than 20 years old. Noma, according to MSF, is so neglected it is not even classified as a neglected disease. This limits the attention Noma gets on the global stage. Little is known about Noma, and the scholarly output as measured by SciVal is less than half (Fig. 17) of the scholarly output of Dracunculiasis, the lowest on the scholarly output ranking (Fig. 3).

**Fig. 17:** Scholarly Output for Noma Disease from 2017 to 2022



So far, MSF has been the biggest advocate for Noma, publishing the most research on the subject and including it in their 2021 report on NTDs, *Overcoming neglect: Finding ways to manage and control NTDs* [41]. MSF, as influential as they are, needs support to bring funding power and to mobilize research capacity.

## Conclusions

My findings are not surprising, and they confirm existing literature: international collaboration leads to favourable outcomes in the form of citation count and then ultimately will lead to favourable outcomes in the form of disease reduction. Predictably, high-income countries produce the most research on NTDs, followed by middle income countries.

High mortality diseases have historically been more researched, and this trend is reflected in NTD research as well. Diseases that have distinct symptoms and are dramatic in presentation are also more researched. In the future, more emphasis needs to be placed on diseases with high morbidity, because they are not as widely researched.

In the future, facilitating international collaboration on NTDs between middle income countries and high- and low-income countries would be very valuable. Considering Brazil's research output in particular and the research capacities of upper-middle income countries, collaboration with Brazilian stakeholders would be beneficial for future research on neglected diseases.

The trajectory of research output and international collaboration on NTD research is very promising. There are concrete goals being set by NGOs and IGOs like the WHO, and there is evidence that those goals are going to be met. More research is being published, indicating

interest and funding is increasing in the realm of NTDs as well. However, there are still NTDs that are wreaking havoc on the world's most vulnerable populations, for example Dengue and Chagas Disease. More international collaboration is necessary on these diseases to assess commonalities between endemic countries and possible solutions outside of the standard MDA programs.

Though significant progress has been made combatting NTDs over the past decade, there are diseases like Noma that are so neglected that they are not even classified as neglected. There needs to be an increase in the dialogue surrounding the list of NTDs between NGOs providing care and the WHO. It is time that the list is updated to bring attention to emerging and existing diseases that are affecting the most vulnerable.

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