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## Coherence between research output and public health priorities: the case of neglected tropical diseases in Brazil<sup>1</sup>

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### Introduction

The gap between health research needs and the research that is funded and undertaken has been calling for managed approaches to the allocation of scarce resources (Røttingen et al., 2013). Priority setting is essential for cost-effective research investment that maximizes health benefits and promotes equity. An increasing number of developing countries have introduced priority-setting processes to identify their most important health problems for which research might provide solutions (Nuyens, 2007). The trend is towards a more rational approach, in which information on what is prioritized and what is actually achieved through research has become crucial.

It has been suggested that diseases that impose a high burden should ideally receive more attention from both funders and the research community (Evans, Shim, & Ioannidis, 2014; Kinge, Roxrud, Vollset, Skirbekk, & Røttingen, 2014). Imbalances between the burden of disease and the research output could indicate diseases that were relatively under- or overrepresented compared to their attributable burden (Albarqouni, Elessi, & Abu-Rmeileh, 2018). Additionally, to foster coherence between research and priorities it is necessary to better understand the current state of the science (the “supply”) and what is required to achieve social goals (the “demand”) (Sarewitz & Pielke, 2007; Wallace & Rafols, 2015).

The high burden of neglected tropical diseases (NTDs) in Brazil called for a better coordination among research and development (R&D) activities and public health programs (Hotez & Fujiwara, 2014). The country has gone through a participatory process to define a National Agenda of Priorities in Health Research (NAPHR) and seven NTDs were included: tuberculosis, Chagas' disease, leprosy, malaria, leishmaniasis, dengue, and schistosomiasis (Ministry of Health, 2006). A solid R&D funding process has been established and a broad health research portfolio was built. The effectiveness of this mechanism in bridging knowledge generation to disease control and reduction of burden still needs to be evaluated.

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This paper focused on assessing the coherence of NTD research output in relation to the established NAPHR and funding investments. The following questions were addressed: i) Was the research reported in scientific publications compatible with the scope of priorities established by the NAPHR (alignment)?; and ii) Are funding and research publications compatible with or proportional to disease burden (consistency)?

The article contributes to the ongoing discussion on research evaluation, policy priorities, agenda setting and NTD elimination, in alignment with the United Nations sustainable development goals (SDG 2030). The assessment of coherence between health research promotion and public health needs serves as an important guidance for funders, donors and scientific community.

## Methods

Three dimensions oriented the analysis: i) Research priorities on NTDs; ii) NTD research output; and iii) Funding for NTD research. The information was used to match the research priorities, publications, and funding, looking at alignment and consistency with disease burden.

### *Research priorities on NTDs*

NTD-related research priorities were retrieved from the NAPHR and each priority was manually categorized according to the type of research that would be needed to address them. The categories were based on typologies of research areas defined for the World Health Organization (WHO) (Terry & van der Rijt, 2010) and the NIH pipeline (Kleinman & Mold, 2009). This approach was taken to develop a common framework to relate research priorities to publications. The categorization was done independently by two authors, discordant classifications were reviewed together and a final classification agreed by consensus. In a few cases, priority lines were assigned to more than one category to account for the different research types that could be used to address them. Six major categories were identified: basic science research; clinical research; population-based research; discovery and development research; health policy and systems research; social sciences research (Kleinman & Mold, 2009; Terry & van der Rijt, 2010).

### *NTD research output*

Data from NTD-related scientific publications were retrieved from Scopus and SciELO (Scientific Electronic Library Online) databases, for the period 2012-2016. The identification of national research was based on articles with at least one author affiliation to a Brazilian institution. Search terms for each of the seven priority NTDs were defined after discussions with experts and validated in the databases.

Publications retrieved in Scopus were matched to the National Library of Medicine's Medical Subject Headings (MeSH) from PubMed. The combination of information from these two databases allowed the use of author affiliations available in Scopus and the MeSH classifications offered by PubMed. An average of 73% of publications was recovered in PubMed across all diseases. The MeSH classification was used as "guidance" for a manual review of all publication titles, including the ones retrieved from SciELO, to assign articles in one of the six major categories previously described. Three parameters oriented this process: i) the research question/problem or objective; ii) "product/result" delivered; and iii) study sample used. Abstracts were assessed whenever one of the three parameters was not obvious in MeSH terms or titles. The categorization was cross-checked among members of the

team, discordant classifications were reviewed together and a final classification agreed by consensus. Only original articles with primary data were categorized.

#### *Funding for NTD research*

The Health Science and Technology Information System, through the “Pesquisa Saúde” database, was used to retrieve information on the amount of funding directed to NTD research by the MoH. The complete database for the period 2012-2016 was retrieved and the following information was extracted for each project: title, abstract, keywords, principal investigator, year and resources granted. NTD-related projects were identified by searching titles and abstracts for the same terms used in publication queries.

#### *Analysis*

Alignment between priorities and output was assessed by comparing their frequency distribution into the six research categories, for each disease separately. Consistency between funding and NTD burden was assessed by looking at the amount and proportion of funding allocated to each disease, as compared to the average burden estimates as reported by the Global Burden of Disease Study (GBD, 2017) for the years 2012-2016. Consistency between the volume of research publications and disease burden was evaluated by looking at the number and proportion of articles in relation to the disease burden. A positive (research surplus) or negative (research deficit) difference between observed and expected number of published articles may represent an unbalanced of the research effort vis-a-vis the relative disease burden (Albarqouni et al., 2018).

## **Results**

#### *Research priorities on NTDs*

Table 1 shows the proportional distribution of priority lines by category for each disease. Priority profiles varied according to disease. Discovery and development research accounted for 43% of malaria priority lines and 38% of schistosomiasis. One-third of tuberculosis priorities were categorized as clinical research, while 31% of dengue priorities were categorized as population-based research. Chagas’ disease research priorities were evenly distributed across basic science, population-based and health policy and systems research. Basic science and social sciences were priorities less common for NTDs in general.

Table 1: Proportional distribution of research priority lines by category for each disease

Priorities	Categories					
Disease	Basic science	Clinical	Population-based	Discovery and development	Health policy and systems	Social sciences
Leprosy	3%	25%	23%	5%	25%	20%
Malaria	0%	14%	14%	43%	29%	0%
Leishmaniasis	11%	6%	22%	28%	33%	0%
Chagas’ disease	33%	0%	33%	0%	33%	0%
Dengue	13%	13%	31%	25%	19%	0%
Schistosomiasis	0%	13%	25%	38%	13%	13%
Tuberculosis	0%	33%	11%	22%	22%	11%

*NTD research output*

The search retrieved a total of 4,817 publications, distributed among the seven NTDs as shown in Figure 1. Leishmaniasis had the highest number of publications with 1,623 articles, followed by tuberculosis (n=1,112), malaria (n=585), dengue (n=560), leprosy (n=440), Chagas' disease (n=311) and schistosomiasis (n=272). Publications were categorized according to the research types addressed (Table 2).

Figure 1: Number of NTDs publications by authors affiliated to Brazilian institutions retrieved from Scopus and SciELO (2012-2016).

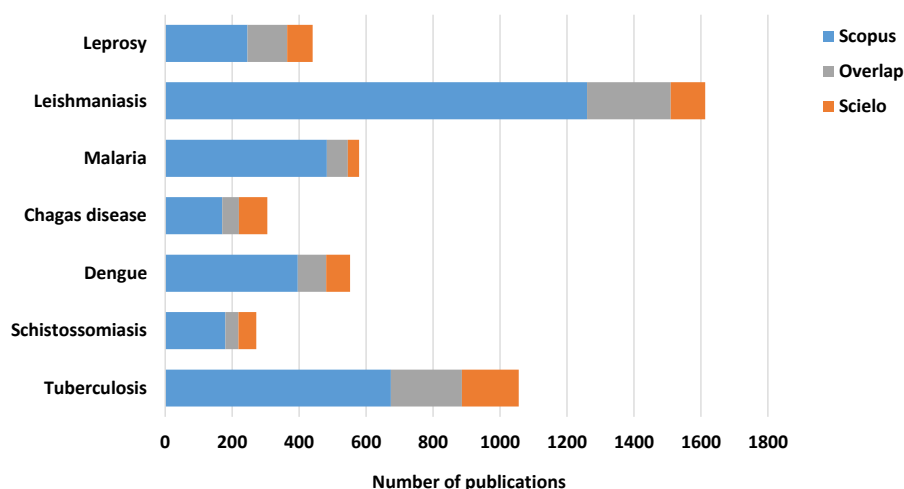


Table 1: Proportional distribution of NTD publications by category for each disease

Publications	Category						
	Disease	Basic science	Clinical	Population-based	Discovery and development	Health policy and systems	Social sciences
Leprosy		22%	31%	25%	7%	6%	8%
Malaria		42%	11%	13%	29%	4%	1%
Leishmaniasis		41%	9%	15%	33%	1%	0%
Chagas' disease		24%	36%	15%	20%	5%	0%
Dengue		29%	17%	25%	14%	9%	5%
Schistosomiasis		31%	18%	19%	30%	2%	0%
Tuberculosis		18%	20%	17%	20%	21%	4%

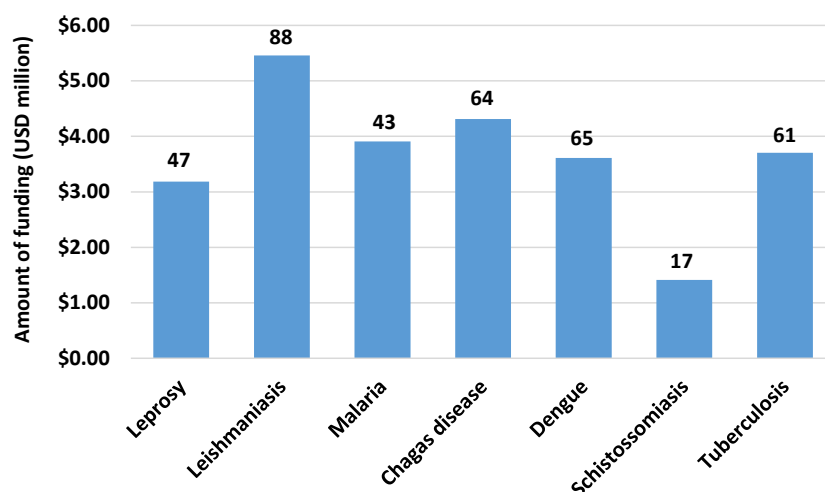
The distribution of publications by category varied greatly between diseases reflecting, to some extent, the capacity and interest of researchers. Basic science research was most frequent in malaria and leishmaniasis publications, with approximately 40% of articles.

Clinical research on leprosy and Chagas' disease accounted for over 30% of publications. Schistosomiasis publications equally emphasized basic science and discovery and development research. Basic science and population-based research accounted for around a third of dengue publications. Twenty-one percent of tuberculosis articles addressed health policy and systems studies. Social sciences and health policy and systems research were the least frequent types of research publications accounting for 0-8% of articles.

#### *Funding for NTD research*

The MoH R&D funding commitment was responsible for a significant expansion of local research capacity to address NTD challenges. Figure 2 displays the funds allocated to each disease during the study period.

Figure 2: MoH research funding for NTDs (2012-2016). Reported values in Brazilian reais were converted to US dollars using the average exchange rate for each year. The number of projects funded for each NTD is indicated in the bars.



Leishmaniasis research received the largest amount of funding in the five-year period, over USD 5 million in 88 projects. Chagas' disease and malaria-related projects had the second and third largest amounts, with approximately 4,3 and 3,9 USD million, respectively. The highest funding level per project granted was on malaria and schistosomiasis, with an average of USD 90,908 and USD 83,219, respectively.

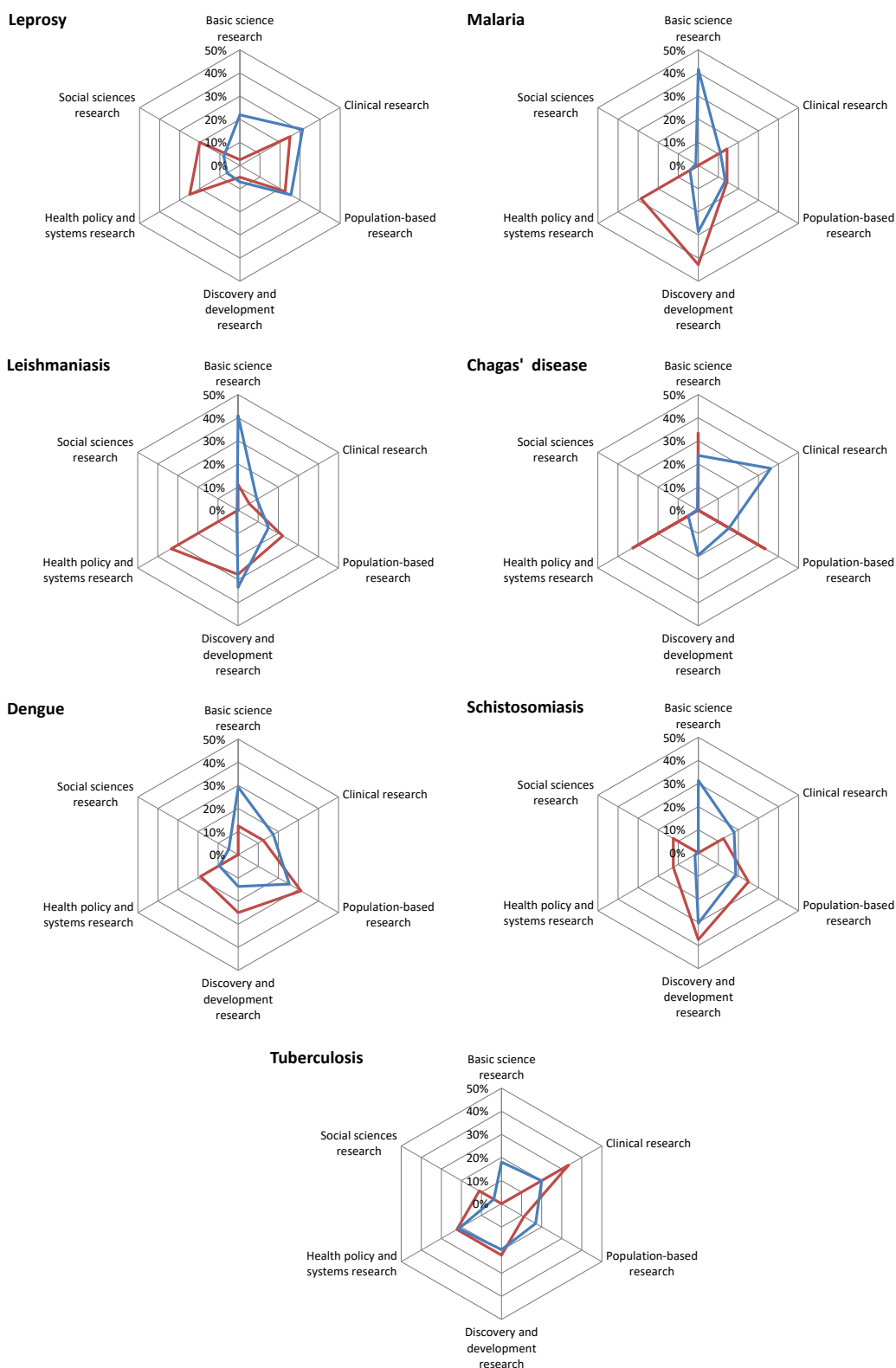
#### *Analysis: matching research agenda, investment, and output*

The matching of NTD priorities and publications suggested only a partial alignment, with the best fit for tuberculosis research, as visualized by the proportional distributions in Figure 3. Health policy and systems research-related priorities were usually unattended by NTD publications, with the exception of tuberculosis. A larger proportion of publications in leishmaniasis, malaria, and schistosomiasis were focused on basic science research. In Chagas' disease, there has been an emphasis on clinical research and discovery and development publications, areas not envisaged in the priority agenda.

A good matching of the priority agenda and publications was seen for clinical research in leprosy; basic science research in Chagas' disease; discovery and development research for malaria, schistosomiasis, tuberculosis and leishmaniasis; population-based research in dengue

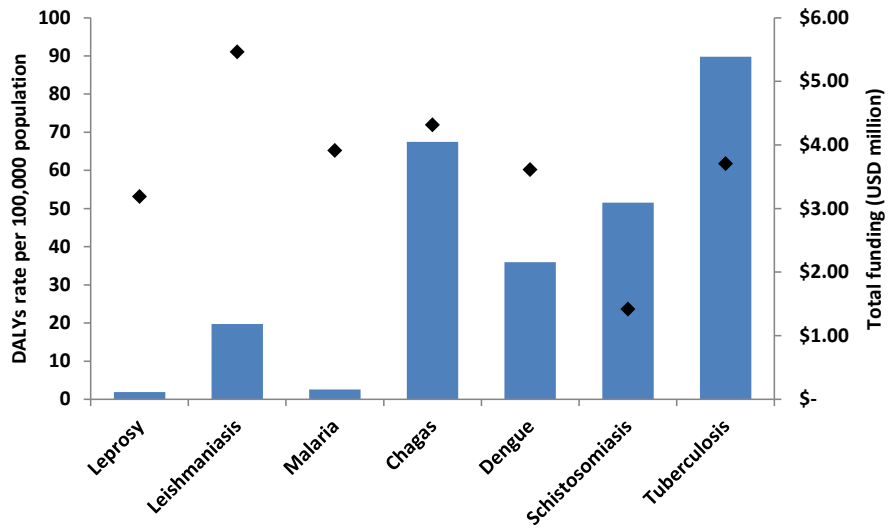
and tuberculosis; and, health policy and systems research in tuberculosis tuberculosis, representing research to address knowledge gaps and disease control needs.

Figure 3: Alignment of research priorities and publications on NTDs. Blue lines indicate publications' and red lines indicate priorities' categories.



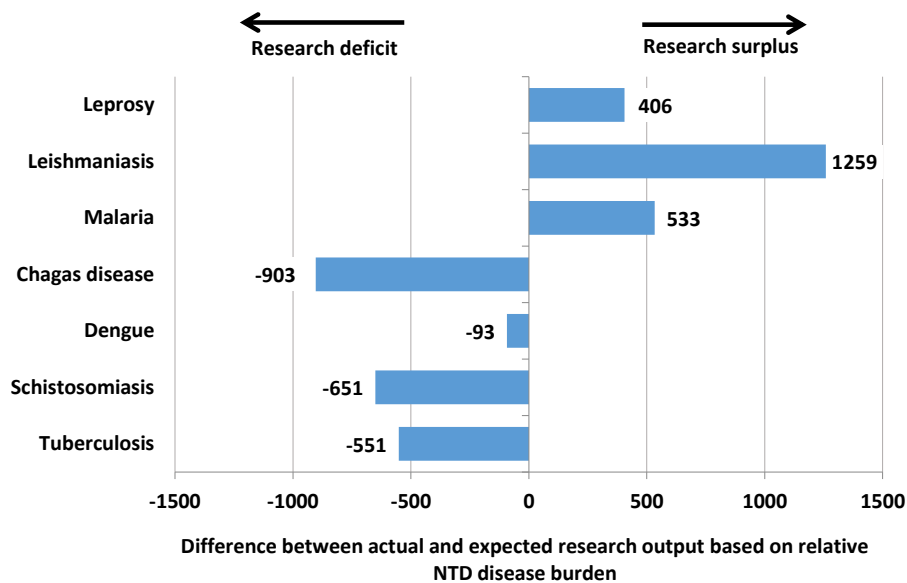
The disease burden, depicted in DALYs rate per 100,000 population, for each NTD is shown in Figure 4. Tuberculosis, Chagas' disease, and schistosomiasis accounted for the highest burden rates with 89.8, 67.5 and 51.6 DALYs/100,000 population, respectively. There was no direct association between level of research funding and burden of the disease: leprosy, leishmaniasis, and malaria with a relative lower burden had higher funding levels, while, the reverse was observed for tuberculosis and schistosomiasis.

Figure 4: Average Disability-Adjusted Life Years (DALYs) rate and funding allocated for NTDs (2012-2016). Bar columns represent DALYs rate per 100,000 population and black diamonds indicate the funding amount.



To further assess the consistency between the volume of research output and disease burden, the difference between observed and expected number of publications were plotted in Figure 5.

Figure 1: Observed and expected number of NTD research articles.



Chagas' disease, schistosomiasis, and tuberculosis showed a research deficit relative to their proportional burden, while leishmaniasis, malaria, and leprosy showed a research surplus. The volume of research was consistent with funding. Leishmaniasis, leprosy, and malaria received the largest amount of funding and had an increased volume of publications relative to their burden. Tuberculosis and schistosomiasis showed the opposite: smaller funding and research deficit.

## **Discussion**

The growing acknowledgment of the role that research plays as a catalyst for socioeconomic development has led to increased demands for monitoring and evaluation of national research systems. Coherence between research agenda, investment and output is, therefore, an important indicator of the functionality of a research system. The data presented herein suggested (i) a partial alignment between national NTD research priorities and scientific publications; (ii) a lack of consistency between disease burden and funding levels, which was also seen between disease burden and volume of publications.

A previous evaluation of the NAPHR fulfillment, although somehow limited, showed that funding allocation was in accordance with health needs (Pacheco Santos et al., 2011). However, an evaluation of whether the outputs of these projects were coherent with priorities has not been carried out. The main criticisms raised by the NAPHR are based on its extensiveness (823 research lines) and difficulty in effectively prioritizing health problems. The good alignment between tuberculosis research and the NAPHR could be related to the close collaboration between the Brazilian TB Research Network (Rede TB) and the National Control Program (Kritski et al., 2016).

Health policy and systems research capacity is generally limited in developing countries. A survey by the Alliance for Health Policy & Systems Research has shown weak institutional capacity and lack of critical mass in most institutions based in these countries (Gonzalez Block & Mills, 2003). In contrast, it has been shown that basic science research play important roles in the Brazilian scientific output (Glänzel, Leta, & Thijs, 2006). Although this type of research is usually dependent on expensive infrastructure and specialized human resources, this could be a reflection of domestic S&T activities and training being modeled on developed countries' standards.

Theoretically, the distribution of research funding for different NTDs should be equitable with respect to the societal burden each type of disease imposes. However, there is not necessarily a linear relationship between burden and funding, because burden of disease is just one of the factors that determine health R&D need (Viergever, 2013). The mismatch between funding and burden of NTDs has been previously described, in a global perspective. Research funding was highly concentrated and had little correlation with the burden of disease, as measured by DALYs (Moran et al., 2009). In many developing countries, the research agenda is often dependent on or guided by external sources of funding, which may divert research efforts away from national interests. It has been acknowledged that priorities of funding agencies may have an important influence on research topics addressed by scientists (Fonseca, Albuquerque, Noyons, & Zicker, 2018).

Alignment and consistency analysis revealed a challenging scenario, especially for leishmaniasis and tuberculosis. The two diseases had strikingly different levels of alignment with the NAPHR and, while the mismatch between publications and priorities was evident for

leishmaniasis, the alignment in tuberculosis was strong. The gap between priorities and research output may be due to several reasons, including the lack of a systematic mapping health R&D needs, difficulties in coordinating a response to established priorities without market influences and other political and socio-economic factors (Viergever, 2013).

It is important to mention that the data used herein was restricted to scientific publications in peer-reviewed journals; therefore, other ways of disseminating research results or unpublished studies were not taken into account. Some mismatch may have occurred in the categorization process, considering that the information collected from publications might not necessarily relate to the priorities set by the NAPHR. As expected, the proposed categories are not totally independent of each other and some articles may fit in one or another group or cover more than one perspective. The funding reported is known to have accounted for a large proportion of the national public sources for NTD research, however, international sources and direct funding from universities and research institutions were not included.

## Conclusions

This study highlights the relevance of monitoring health needs, research agendas, investment and research outputs to optimize the uptake of scientific evidence into health, particularly in developing countries, where resources are scarce and research capacity limited. Our results show that the analysis of the NTD research portfolio could help identify research topics and promote research more closely aligned to social needs. The triangulation of priorities, output and funding data results offered a stronger perspective with potential to inform policy decision in public health. A comprehensive and dynamic national research information system could offer quality data for better assessment of the return and impact of the scientific activity in the country.

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