

Research scenario in physiotherapy for people with *human t-cell lymphotropic virus (HTLV)*: scientometric study

Cenário das pesquisas em Fisioterapia para pessoas com *Human T-Cell Lymphotropic Virus (HTLV)*: estudo cientométrico

DOI:10.34117/bjdv8n2-238

Recebimento dos originais: 07/01/2022

Aceitação para publicação:16/02/2022

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ABSTRACT

Introduction: *Human T-Cell Lymphotropic Virus (HTLV)* is a sexually transmitted retrovirus that can cause chronic pain of moderate to high intensity, spasticity and weakness in the lower limbs, balance, gait, urinary and sexual dysfunctions. Physiotherapy is a treatment that has been shown to be effective in improving symptoms and signs. However, research on the topic is scarce. **Objective:** Outline the research scenario in Physiotherapy aimed at people infected with HTLV-1. **Methods:** Scientometric study carried through Medline, Virtual Health Library, Scopus, Web of Science and Google Scholar databases. The metadata regarding the studies were tabulated and analyzed using descriptive statistics. The VOS.Viewer software was used for the analysis of quotes. Clinical studies were also analyzed considering meta-scientific impacts including analysis of conflicts of interest and methodological biases. **Results:** Initially 1160 studies were identified. After applying the eligibility criteria, 68 articles remained, of which 34 articles were observational studies, 21 on intervention, 9 on review, two on qualitative research and two on diagnostic accuracy. Brazil is responsible for 94.11% of the production on the theme. The sub-areas of Physiotherapy covered by

the studies were neurofunctional and urogenital, using different exercise modalities, proprioceptive neuromuscular facilitation, biofeedback and non-invasive neuromodulation. **Conclusion:** On the topic of Physiotherapy for people with HTLV-1, Brazil occupies a prominent place in scientific production. Research on the subject receives little or no funding, requiring free of charge publications in journals, which affects its visibility.

Keywords: Physiotherapy, *Lymphotropic Virus T Type 1 Human*, Tropical Spastic Paraparesis, Science.

RESUMO

Introdução: *O vírus linfotrófico de células T humanas (HTLV) é um retrovírus de transmissão sexual que pode causar dor crônica de moderada a alta intensidade, espasticidade e fraqueza nos membros inferiores, equilíbrio, marcha, disfunções urinárias e sexuais. A fisioterapia é um tratamento que tem se mostrado eficaz na melhoria dos sintomas e sinais. No entanto, as pesquisas sobre o tema são escassas.* **Objetivo:** Descrever o cenário da pesquisa em Fisioterapia voltada para pessoas infectadas pelo HTLV-1. **Métodos:** Estudo cientométrico realizado através do Medline, Biblioteca Virtual em Saúde, Scopus, Web of Science e bancos de dados de bolsistas do Google. Os metadados referentes aos estudos foram tabulados e analisados utilizando estatísticas descritivas. O software VOS.Viewer foi utilizado para a análise de citações. Os estudos clínicos também foram analisados considerando os impactos meta-científicos, incluindo a análise de conflitos de interesse e de viés metodológico. **Resultados:** Inicialmente foram identificados 1160 estudos. Após a aplicação dos critérios de elegibilidade, restaram 68 artigos, dos quais 34 eram estudos observacionais, 21 sobre intervenção, 9 sobre revisão, dois sobre pesquisa qualitativa e dois sobre precisão diagnóstica. O Brasil é responsável por 94,11% da produção sobre o tema. As sub-áreas de Fisioterapia cobertas pelos estudos foram neurofuncionais e urogenitais, utilizando diferentes modalidades de exercício, facilitação neuromuscular proprioceptiva, biofeedback e neuromodulação não-invasiva. **Conclusão:** Sobre o tema Fisioterapia para pessoas com HTLV-1, o Brasil ocupa um lugar de destaque na produção científica. A pesquisa sobre o tema recebe pouco ou nenhum financiamento, exigindo publicações gratuitas em periódicos, o que afeta sua visibilidade.

Palavras-chave: Fisioterapia, *Vírus Lymphotropic T Tipo 1 Humano*, Paraparesia Espástica Tropical, Ciência.

1 INTRODUCTION

The Human T-Cell Lymphotropic Virus (HTLV) is a sexually transmitted retrovirus that presents increasing contamination due to the vast majority of those infected being asymptomatic and transmitting it silently and unconsciously^{1,2}. However, about 5% of those infected have their health impaired with losses in quality-of-life³, functional disabilities⁴, dependence for carrying out activities of daily living⁵ and a consequent reduction in social participation.

Among the main diseases caused by HTLV, especially type 1 (HTLV-1), about 1.8% of those infected develop myelopathy associated with HTLV-1 also known as tropical spastic paraparesis or HAM/TSP². HAM/TSP occurs due to viral injury of the thoracolumbar region of the spinal cord⁶. This myelopathy causes chronic pain of moderate to high intensity, spasticity and weakness in the lower limbs, affects balance and gait, and generates sexual and sphincter dysfunction^{7,8}. It is an incurable disease due to the lack of vaccines and the ineffectiveness of antivirals, which requires therapeutic actions to relief signs and symptoms. Physiotherapy presents itself as a relevant therapeutic alternative for using low-cost physical resources that improve movement, balance, gait and sexual and sphincter functions, aside from providing pain relief⁹.

However, HTLV is a condition that mainly affects a population of low socioeconomic status¹⁰ and receives a low volume of funding for research, especially when compared to the resources allocated to the studies of other retroviruses such as HIV-AIDS¹¹. Added to this reality is the fact that Physiotherapy is also a sub-area of health that receives a very low level of funding for conducting research¹². The existence of few resources affects the development of science in neglected areas and themes. Scientometric studies can help unravel the aspects that cross research in Physiotherapy for people with HTLV-1 and generate data for their support. Scientometrics is a research modality that analyzes the metadata of studies published in specific topics and areas¹³.

Data are extracted from the publications with or without the aid of search systems and analysis of citations and networks, which allow quantitative and qualitative analysis using statistical tools and theoretical foundation of meta-science¹⁴. Few scientometric studies were carried out to analyze Physiotherapy, and these focused more on the profile of research physiotherapists^{15,16}. However, this method of investigation can help to identify gaps to be filled in future research and to understand phenomena that reduce the visibility of scientific findings on the subject. The objective of this work is to outline the scenario of research in Physiotherapy aimed at people infected with HTLV-1.

2 METHODS

STUDY DESIGN

Scientometric study performed in the following databases: Pubmed, Medline, Lilacs, SciELO, Bireme, Web of Science, Scopus and Google Scholar.

CRITERIA OF ELIGIBILITY

We searched for works on physiotherapeutic treatment or general studies in the large area of health in which physiotherapists participated in the HTLV-1 theme. Clinical studies of intervention (protocols, pilots, case studies, case series, single-arm, randomized and crossover clinical trials), observational (cross-sectional and longitudinal), diagnostic accuracy, validation of instruments and mechanisms of actions of physical therapy resources. In addition to these, interdisciplinary studies with the participation of physical therapists were included, such as epidemiology studies, narrative and systematic reviews, among others, as long as they are applicable to research in Physiotherapy on the subject of HTLV. Those who addressed the exclusivity issue of another health sub-area such as involving the effect of drugs, studies on vaccines, cellular and molecular studies, with experimental animals, with children and those who were not in the following languages were excluded: English, Spanish and Portuguese, or those with no possible access to it. The collection included studies published until December 2020.

SEARCH STRATEGY

The search strategy used in Pubmed was: ("physical therapy modalities"[MeSH Terms] OR ("physical"[All Fields] AND "therapy"[All Fields] AND "modalities"[All Fields]) OR "physical therapy modalities "[All Fields] OR "physiotherapy"[All Fields]) AND ("human t-lymphotropic virus 1"[MeSH Terms] OR "human t-lymphotropic virus 1"[All Fields] OR "htlv 1"[All Fields]). In the other databases, the same terms were used interspersed with Boolean operators AND, NOT and OR.

COLLECTION PROCEDURE

The collection was carried out by two researchers independently and dissents were resolved through agreement. The selected metadata were entered into a previously prepared database.

STUDY VARIABLES

From the studies, were analyzed the most used methodological designs, the area of concentration, the most used periodicals for publications, the impact factor of these periodicals, the amount charged for the publication, the mode of access, the year and the place of publication.

From the authors, the profession, degree, linking institutions, funding sources, degree, total number of publications, number of publications on the HTLV and the H index of the main researchers (first, second-last and last author) were analyzed.

From the meta-scientific information of the intervention studies, the themes of the studies, the main result (classified as positive or negative), the primary and secondary outcomes, the studied therapeutic modalities and the sub-area study were analyzed.

DATA ANALYSIS

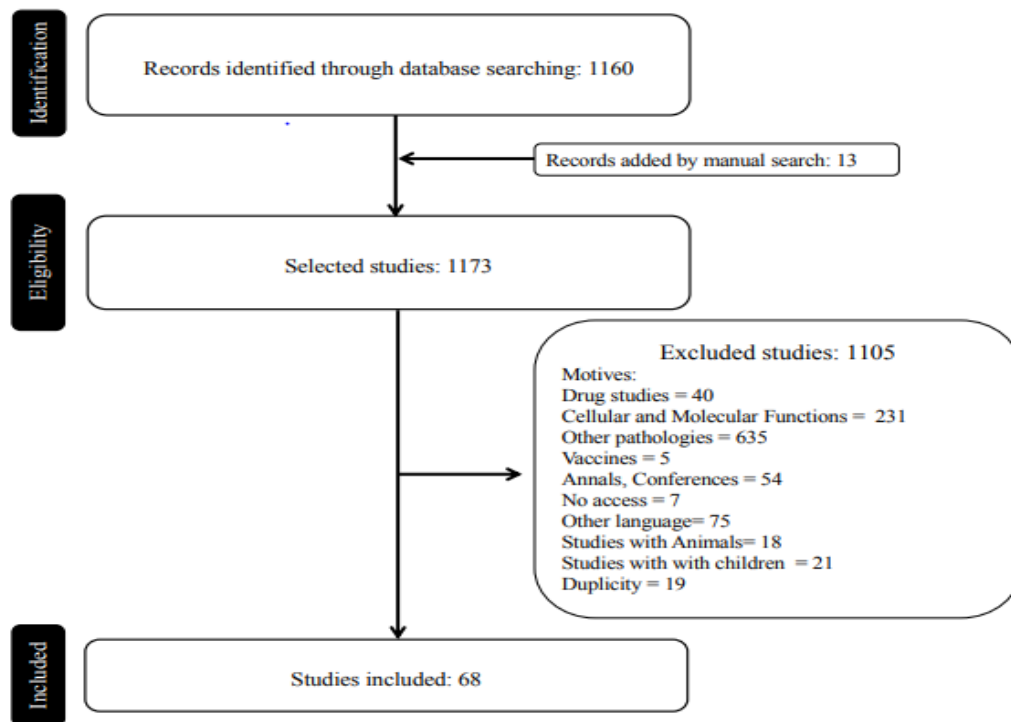
Data were tabulated in a database and analyzed in the Statistic Package for Social Science (SPSS) version 14.0 through descriptive and inferential analyses.

For the construction of co-authorship networks, the Vosviewer software was used, using the Full Accounting counting method. The size of each node is determined by the weight of each one of them, in the case of co-authorship, the number of articles published by an author was analyzed. The edge, line connecting the nodes, represents the joint publication of two authors. The node color can represent a cluster to which the author belongs or the years of the works publications.

3 RESULTS

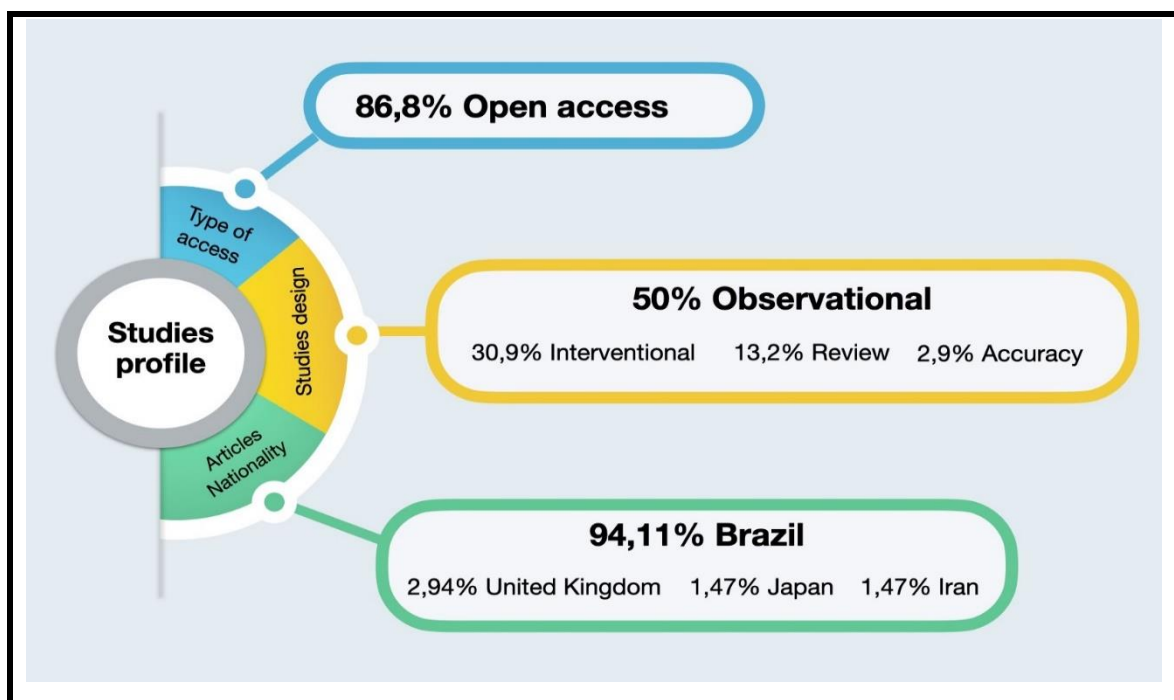
The preliminary search identified 1173 articles. After reading the titles and abstracts, 1105 of those were excluded, leaving 68 studies that followed for analysis. The reasons for the exclusions can be seen in figure 1.

Figure 1: Flow chart of data collect



Of the 68 that were included, 34 (50%) were observational studies, 21 (30.9%) were intervention studies, and 9 (13.2%) were review studies. The type of open access was the most common (86.80%) and Brazil is responsible for 94.11% of publications on the subject (Figure 2).

Figure 2. Descriptive analysis of the selected studies profile involving HTLV-1 Physiotherapy.



Regarding the methodological designs, half are observational studies, of which 30 (44.1%) are cross-sectional, seven (10.3%) are narrative reviews, six (6.8%) are randomized clinical trials and the others distributed between qualitative research, diagnostic accuracy, cases or pilots (Table 1).

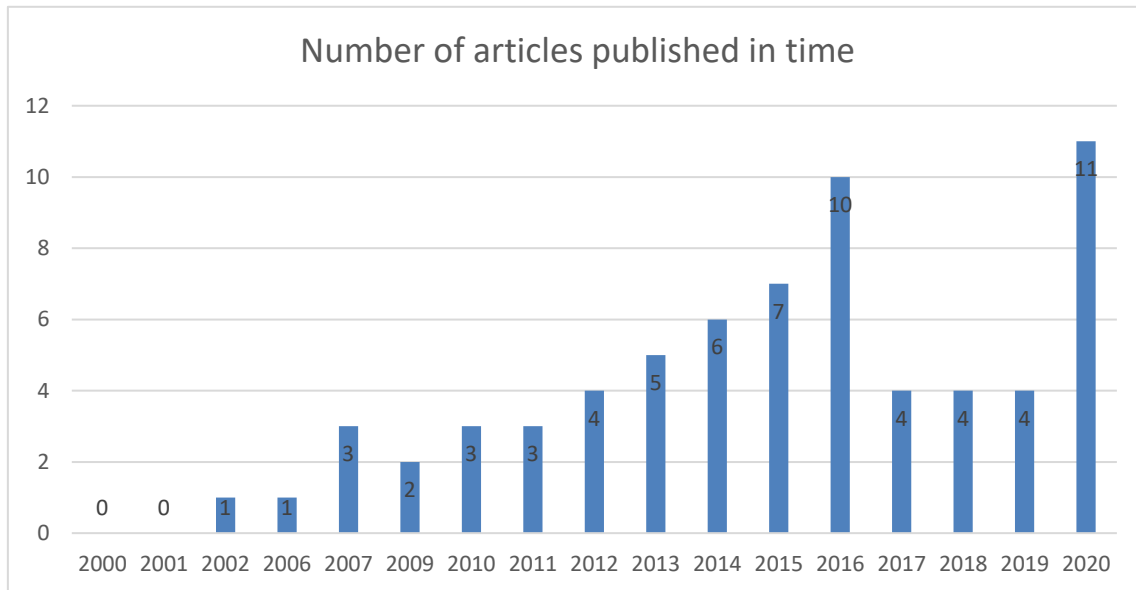
Table 1. Distribution of selected study designs involving HTLV-1 Physiotherapy.

Study Design	N	%
Observational		
Cross Seccional	30	44.1
Cohort	4	5.9
Reviews		
Narrative	7	10.3
Systematic	2	2.9
Interventions		
Unmanaged Clinical Trial	5	7.4
Randomized Clinical Trial	6	8.8
Crossed Clinical Trial	2	2.9
Pilot Study	4	5.9
Case Study	4	5.9
Qualitative Research		
Interview and Focus Group	2	2.9
Diagnostic Accuracy		
Biomechanical Diagnosis	2	2.9

As for the distribution of studies by countries, it was observed that 64 (94.11%) were obtained in Brazil, two (2.94%) in the United Kingdom, one (1.47%) in Japan and one (1.47 %) in Iran. Looking specifically at Brazil, we found that Bahia is the place where most of the studies on the subject were produced, accounting for 34 (50%) of the publications, but 13 (19.11%) studies were also identified in Pará, 11 (16.17%) in Rio de Janeiro, three (4.41%) in Minas Gerais, one (1.47%) in the states of São Paulo, Mato Grosso do Sul and Paraná. These properties are authorized to 28 (41.17%) public institutions and 36 (52.94%) private institutions, and it is not possible to identify the profile in four (5.88%) institutions.

The journals had the registration code in the International Standard Serial Number (ISSN) in 98.52% of the cases and registration in the Digital Object Identifier System (DOI) in 60 (88.2%) of the published manuscripts. Twenty-four (35.3%) articles were selected from the Pubmed database, 20 (29.4%) from SciELO and 9 (13.2%) from LILACS. The distribution at the time of the publication of these articles can be seen in Figure 3.

Figure 3. Distribution of HTLV Physical Therapy Publications on Time.



Regarding the authors, at the time of publication, 68 (22.51%) had a PhD degree, 70 (23.17%) a master's degree, 27 (8.94%) were undergraduate students, 13 (4.30%) were graduates and 23 (7.61%) were specialists. Among the authors, it was observed that the predominant profession was physiotherapists or physiotherapy students, with 196 (65.11%) authors, but there were also 69 (22.92%) physicians, five (1.66%) pharmacists, four (1.32%) nurses and biomedical doctors, three (0.99%) biologists, physical educators and psychologists, two (0.66%) dance teachers, dentists, statisticians and sociologists, and one (0.33%) occupational therapist.

Image 4a. Co-authorship Analysis on the Physiotherapy and HTLV topic.

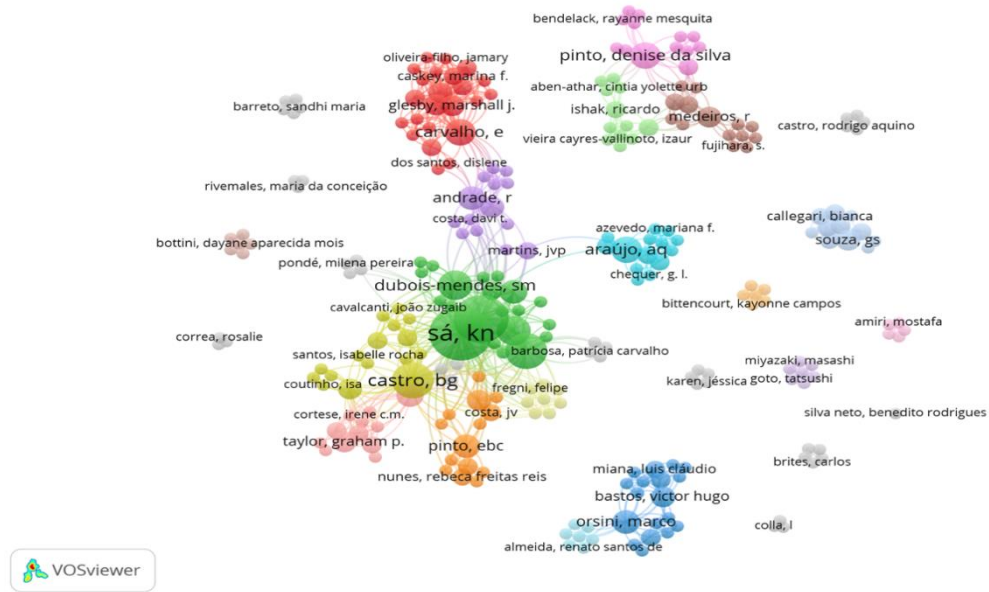
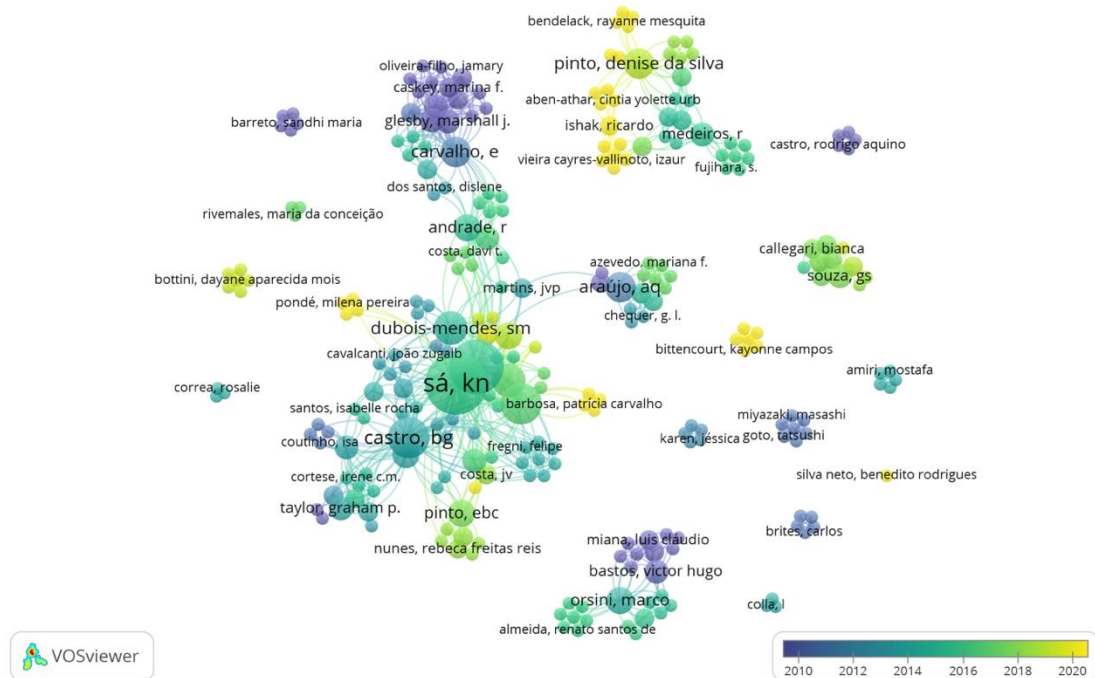


Image 4b. Analysis of Co-authorship in Physiotherapy and HTLV, stratified in the last 10 years of publication.



It can be observed in Image 4a that, despite the existence of several isolated researchers groups (Amiri, Brites, Castro, Bittencourt, Bottini, Correa, Callegari, Karen, Silva Neto and Colla) researching the topic of Physiotherapy at HTLV, there is a network of consolidated collaboration involving groups of researchers in Bahia (Sá, Mendes, Castro, Carvalho, Andrade, Pinto, Cavalcanti, Nunes), Rio de Janeiro (Araújo, Checker

and Azevedo), São Paulo (Baptista), United Kingdom (Taylor and Martins) and the United States (Fregni). In the last 10 years, peaks of scientific production on the subject have been observed in 2016 and 2020. In 2016, the co-authorship evidenced in the main collaboration networks is observed. However, in 2020 it is possible to observe the emergence of a new network (Pinto, Ishak, Medeiros, Aben-Athar, Bendelack, Vieira Cayres and Fujiaki) from Pará (Figure 4b). On average, these authors have an h-index on Google Scholar of 27.0.

Regarding the themes of the studies, 48 (70.6%) focused on neurofunctional aspects such as gait, spasticity, balance, posture and pain; while 6 (8.8%) were intended for the treatment of urogenital symptoms, two (2.9%) were for pulmonary symptoms and three (4.4%) dealt with epidemiological and collective health aspects of the subject.

When we analyzed the 21 (30.9%) intervention studies, most were developed with symptomatic people for HAM/TSP and the result for the main outcome was positive in 16 (76.19%) studies, negative in one and in two they did not observe intra- and inter-group differences. The therapeutic modalities applied to the HTLV were: kinesiotherapy (including Pilates exercises, functional exercises, exercises with virtual reality, biofeedback and home exercise programs with the aid of a booklet), proprioceptive neuromuscular facilitation (PNF), transcranial stimulation by direct current (tDCS) and by repetitive magnetic stimulation (rTMS).

As for the analysis of conflicts of interest, 31 (45.58%) works declared no conflict of interest and 37 (54.41%) did not present the declaration. No company-funded studies were identified. In only five (7.35%) there was reference to scholarship funding for one of the authors without any support for the referred research project.

4 DISCUSSION

This scientometric study on the topic of Physiotherapy for HTLV-infected people found publications of observational, intervention, qualitative research, diagnostic accuracy and review studies. Brazil is responsible for most of the scientific production on the subject. The physiotherapy sub-areas covered by the studies were neurofunctional, urogenital and respiratory, using different outcomes and therapeutic modalities.

The open access to information model found in most publications allows physical therapists around the world to base their clinical practice on the evidence shown in the studies. Most studies are published in journals indexed in LILACS and SciELO, although some are only available in Pubmed. Despite the countless efforts of Brazilian researchers

and institutions for the internationalization of Brazilian research, the international visibility of studies indexed in SciELO is still low¹⁷.

The publication of research results in open access has become consensual because it is accountable to society about the resources used in research, which, in general, are public. Most of the periodicals are edited by professional societies, scientific associations or non-profit university institutions, which make use of the contributions of their members and of their own or governmental subsidy to fund the publications. Thanks to the creation of SciELO in 1998, the program is funded by FAPESP (90%) and by CNPq (10%). It is estimated that the average publication cost per article is US\$130.00. The Brazilian Journal of Medical and Biological Research, for example, which was part of the ten pioneering journals that initially formed SciELO, started charging from 2004 the amount of R\$1800.00 (US\$790.00) for articles by national and US\$900.00 for foreign authors. These values are much lower than those charged internationally, which is based on the PLoS ONE group, which charges an average of US\$ 1350.00 to US\$ 2250.00 for moderate impact and renowned journals from US\$ 2700.00 to US\$ 2900.00. These high publication costs reduce the ability to disseminate the knowledge produced¹⁸.

A study carried out in countries of the European community, the United States, Japan, Canada and Brazil concluded that 50% of the articles published between 2004 and 2011 in these places are available in open access¹⁹. Countries that receive little or no funding are less likely to publish in open access journals, forcing the search for scientific journals that do not cover publications, which has been achieved in practically only a few journals indexed in SciELO.

It was not possible to understand the behavior of the scientific production curve on the topic, which presented a linear progressive growth until 2015, but in the period from 2016 to 2019 there was a significant drop in the number of publications on the topic. Future studies should investigate what happens in periods of decline in the number of publications.

Although most studies are observational, the scientific production on the subject includes clinical studies that can support the choice of physical therapy modalities. It is common for science to start understanding a phenomenon in the observational model to then generate hypotheses for interventions that can be used in the treatment of observed dysfunctions and test them in clinical studies²⁰.

Based on the observational studies, it is confirmed that the main problems that affect these patients involve disorders of gait^{21,22,8,23}, balance^{24,25,26}, pelvic floor^{27,28},

spasticity^{29,30} and pain^{10,31,32,33}. These dysfunctions affect daily life activities^{5,34} and quality of life³⁵. These outcomes, therefore, should be systematically evaluated in surveys to monitor the evolution of the disease and the efficacy and safety of clinical interventions. These are outcomes that can benefit from physical therapy intervention.

Brazil occupies a leading role in research in this field. One of the reasons for this finding may be related to the fact that half of the physiotherapists in the world are in the country¹⁶. We hypothesize that most research on HTLV Physiotherapy is in a country where the prevalence of infected people is high. Despite this, HTLV is also a prevalent infection in Japan, Africa and South and Central America. It is especially noteworthy that countries with a high socioeconomic and scientific level such as Japan, France, the United Kingdom and Australia do not present relevant scientific production on the topic of Physiotherapy at HTLV, dedicating themselves more to molecular research, immunological and on drugs and vaccines in the studies developed for this population.

As for the physical therapy resources used in the treatment, the use of individual outpatient kinesiotherapy was identified through functional exercises^{36,37,38,39}, home exercises⁴⁰, proprioceptive neuromuscular facilitation^{41,42}, Pilates^{43,44}, virtual reality^{45,46}, respiratory training⁴⁷, electrical stimulation^{48,39}, neuromodulation with tDCS⁴⁹ and rTMS²⁹. Most of these resources have no side effects, are low cost, help to delay the evolution of dysfunctions and have a positive impact on quality of life. Although there are still a small number of clinical trials with small samples and small effect sizes, Physiotherapy has benefits to treat the main signs and symptoms that affect this population. Many studies are needed that can guarantee an evidence-based practice.

Another study design relevant to strengthening research in an area involves the ability to accurately assess clinical outcome. Two studies of diagnostic accuracy were found. One addressed the use of the International Classification of Functioning (ICF)³⁰ and the other the use of methods to assess biomechanics and postural control in this population⁵⁰. The more accurate the diagnosis, the greater the possibility that research will detect the differences obtained by an intervention and the greater certainty about its effectiveness for clinical use.

Most study authors are physical therapists, followed by physicians and then by other professionals, which confirms the hypothesis that most authors are physical therapists. Despite this, it is necessary to recognize important physicians who guided the training of Brazilian researchers with a degree in Physiotherapy. It is important to point out that the number of physical therapists with a PhD degree is much smaller than that of

physicians. However, the interest of researchers in rare and neglected diseases is low in all professions. Journals that report research on the topic have a low impact factor, which affects the h-index of authors and, consequently, the interest of researchers. Mainly research professors from master's and doctoral programs prefer themes that raise this index, which is an important indicator to be achieved according to the criteria of the regulatory and evaluation bodies. The limited participation of authors with a PhD degree creates a weakness, both in terms of scientific maturity and the low capacity to raise funds for research. Authors' titles are still a relevant factor related to better quality works.

The co-authorship analysis confirms that most publications on the subject occur in Brazil, especially in the states of Bahia, Pará, Rio de Janeiro and Minas Gerais. Isolated initiatives appear in Iran, Japan and the United Kingdom. Cooperation between researchers is a way used to face the low level of funding that these researches receive. There is, however, the need to foster international cooperation.

We emphasize the pioneering nature of this scientometric study on the topic of Physiotherapy at HTLV-1. Because of this, the discussion of the findings was limited. Another important limitation was related to the absence of some metadata in the selected studies, such as titles and profession of the authors and professional data that we can find only for Brazilians through the Lattes platform.

In short, it was possible to identify that there are few works that study Physical Therapy in HTLV-1, mostly observational studies, available in open access journals developed in Brazil, which is an endemic country with low funding for research. There are few doctors dedicated to research on the subject, especially testing the efficacy and safety of physical therapy modalities, which causes low funding, little visibility of this research and low level of evidence for interventions. However, the limited number of interventions that help to reduce the suffering of those affected justifies the support that should be given to research on Physiotherapy to treat people with HTLV.

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