The minimum dataset for rare diseases in Brazil: a systematic review

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Review question

What is the minimum data set used in registries for rare diseases?

Searches

The search strategy aims to find both published and unpublished studies. As recommended by the Joanna Briggs Institute (JBI), a three-step search strategy will be applied. A first limited search in PubMed database will be performed followed by an analysis of the text words contained in the title and abstract, and the index terms available at MeSH (Medical Subject Headings), a vocabulary thesaurus used for indexing articles for PubMed. A second search using all identified keywords and index terms will then be undertaken across all included databases. Lastly, the reference list of all identified reports and articles will be searched for additional studies. Only English and Portuguese language papers, not limited by date, will be included. Six databases were chosen for studies search: PubMed, CINAHL (Cumulative Index to Nursing and Allied Health Literature), Scopus, EBSCO (Health Source - Consumer Edition), MEDLINE and Web of Science.Documents from the World Health Organization (WHO) and governmental websites will also be examined for policies and reports. The search for gray literature studies will include Google Scholar, ProQuest Dissertations and Theses, Open Thesis, BDTD (Brazilian Digital Library of Theses and Dissertations), and NDLTD (Networked Digital Library of Theses and Dissertations). The search terms will be "rare disease", "digital health", "minimum dataset", "national network"and "national plan"

Types of study to be included

Any study design (i.e., quantitative, qualitative, or mixed methods) as well as opinion pieces, commentaries, letters and editorials, will be considered. National plans and policies Industry reports, position papers, or program reports will be included. Review articles will be excluded, but relevant papers will be used to crosscheck for primary papers. Personal blogs and social media posts will be excluded.

Condition or domain being studied

The requirements of data sharing within the rare disease realm are compelling, i.e., simply data exchange is not enough. Our proposal intends to demonstrate that effective data sharing networks require consensus on data syntax, meaning, and security of information in the Brazilian scenario. In this case, the solution must be able to understand and communicate minimally with other systems and need to be according Brazilian Policy of Comprehensive Care for People with Rare Disease and global literature. We expect that the outcomes will bring pieces of evidence about the best practices using the minimum dataset approach for these networks that involve many stakeholders, e.g hospitals, universities, and primary units care can benefit. Our domain will include all rare disease networks in healthcare systems.

Participants/population

The population consists of networks that have their data incorporated within a national or institutional environment that describes minimum datasets for rare disease data sharing.

Intervention(s), exposure(s)

Any network, national plan, or strategy that describes minimum dataset for rare diseases used to facilitate the comprehension of concepts, strategies, methods, or techniques for rare disease field improvement in Brazil.

Comparator(s)/control
No comparator is required.

Context
This review is part of and will support an umbrella project that is a design mixed prospective and retrospective observational cohort study to map the scenario of rare diseases in Brazil.

Main outcome(s)
Some outcomes of interests specific for minimum datasets will involve information about epidemiology, clinical procedures, and therapeutic resources among other features. Besides that, we hope to find standards in socio-demographic, epidemiological, clinical and therapeutics data able to identify and characterizes the type of treatments, the existing diagnostic and technological resources within networks. Thus, to map concerns about human resources to support information about what specialists are required to elaborate a network according to the guidelines of WHO, and the national plans.

* Measures of effect

Not applicable

Additional outcome(s)
- tools and techniques applied results;

- Developments in public health, primary care and hospital processes;

- Research outcomes of rare diseases;

- New approaches for ethical and legal issues.

* Measures of effect

Not applicable

Data extraction (selection and coding)
Data to be extracted will likely include study characteristics such as author(s), publication year, title, country of publication, national context, study objective(s), sample size, data collection method, data analysis method and main findings and strength of evidence recommendation according to the quality assessment tool. Thus, this step of charting will be iterative, enabling the reviewers to update the data extraction form. Two independent reviewers will screen the abstracts and full articles. Divergences between the two reviewers will be discussed. Should reviewers not reach an accord, a third reviewer will be participating in the process to make a final decision on inclusion or exclusion. Reviewers will not be blinded to the journal, title, study authors, or associated institutions.

Risk of bias (quality) assessment
The methodological quality of the included systematic reviews will be assessed by two independent reviewers using the multiple systematic reviews assessment tool (AMSTAR 2). Regarding the quality evaluation of the results and the strength of the evidence recommendations found in our review, the GRADE approach (Grading of Recommendations Assessment, Development and Evaluation) will be adopted.

Strategy for data synthesis
In the results section, data will be visually represented in tables and diagrams using data extracted from the included papers. According to the type of studies, the review output will be presented in a descriptive format that aligns with the objectives and scope of the review. The narrative synthesis will seek to investigate similarities and differences between studies to explore patterns, themes, and relationships and propose explanations for findings, e.g., how and why certain minimum datasets development or implementation strategies have worked, or not, in the rare disease field.

Analysis of subgroups or subsets
None.

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Type and method of review
Epidemiologic, Methodology, Synthesis of qualitative studies, Systematic review

Anticipated or actual start date
05 January 2021

Anticipated completion date
25 March 2021

Funding sources/sponsors
This research is under development and is part of the expected results of The National Network of Rare Diseases project. It's was approved in Edital nº. 25/2019 from CNPq ('Brazilian Council for Scientific and Technological Development, in Portuguese) with financial support from the Ministry of Health, in the amount of 3.5 million reais.

Grant number(s)

State the funder, grant or award number and the date of award

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Conflicts of interest

Language
English, Portuguese-Brazil

Country
Brazil

Stage of review
Review Ongoing

Subject index terms status
Subject indexing assigned by CRD

Subject index terms
MeSH headings have not been applied to this record

Date of registration in PROSPERO
14 February 2021

Date of first submission

14 January 2021

Stage of review at time of this submission

Stage	Started	Completed
Preliminary searches	Yes	No
Piloting of the study selection process	No	No
Formal screening of search results against eligibility criteria	No	No
Data extraction	No	No
Risk of bias (quality) assessment	No	No
Data analysis	No	No

The record owner confirms that the information they have supplied for this submission is accurate and complete and they understand that deliberate provision of inaccurate information or omission of data may be construed as scientific misconduct.

The record owner confirms that they will update the status of the review when it is completed and will add publication details in due course.

Versions

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