INPLASY PROTOCOL

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Support: **None.**

Review Stage at time of this submission: **The review has not yet started.**

Conflicts of interest:
None declared.

Pulmonary function in people with myotonic dystrophy: a systematic review and meta-analysis

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Review question / Objective: **Is pulmonary function reduced in people with myotonic dystrophy compared with their predicted values?**
Condition being studied: **Myotonic dystrophy.**
Study designs to be included: **We will include randomised clinical trials (RCTs), quasi-randomised controlled trials, and observational studies (retrospective, prospective, cross-sectional, longitudinal, case-control, and cohort). All editorials, letters, review articles, systematic review, and meta-analysis and in vitro studies will be excluded.**

INPLASY registration number: This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 23 March 2022 and was last updated on 23 March 2022 (registration number INPLASY202230130).

INTRODUCTION

Review question / Objective: **Is pulmonary function reduced in people with myotonic dystrophy compared with their predicted values?**

Condition being studied: **Myotonic dystrophy.**

METHODS

Participant or population: **Patients with myotonic dystrophy.**

Intervention: **Pulmonary function assessment.**

Comparator: **No applied.**

Study designs to be included: We will include randomised clinical trials (RCTs), quasi-randomised controlled trials, and observational studies (retrospective, prospective, cross-sectional, longitudinal, case-control, and cohort). All editorials, letters, review articles, systematic review, and meta-analysis and in vitro studies will be excluded.

Eligibility criteria: Studies that investigate pulmonary function in people with myotonic dystrophy.

Information sources: We will review the Embase, Cochrane Library, CINAHL, Web of Science, and PubMed/MEDLINE databases.

Main outcome(s): Forced vital capacity (FVC), measured in litres and predicted value.

Additional outcome(s): 1. Forced expiratory volume during the first second (FEV1), measured in litres and predicted value; 2. Maximal inspiratory pressure (MIP), measured in centimetres of water and predicted value; 3. Maximal expiratory pressure (MEP), measured in centimetres of water and predicted value; 4. Peak cough flow (PCF), measured in litres/minute and predicted value.

Quality assessment / Risk of bias analysis: The risk of bias and the quality of observational and interventional studies will be assessed using the corresponding assessment tools recommended by the National Heart, Lung, and Blood Institute (NHBLI). This assessment will be done by two investigators (SY-FL), with disagreements being resolved by arbitration by a third investigator (RTC). The level of confidence in summary effect estimates will be assessed using the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) approach. Publication bias will be assessed from funnel plots.

Strategy of data synthesis: We will conduct the bibliographic search and compile the identified records in Rayyan QCRI (Rayyan QCRI software, Qatar Computing Research Institute (Data Analysis), Doha, Qatar). All studies identified by the search strategy will be screened against the eligibility criteria by two reviewers (AC-FL) working independently based on title and abstract. We will remove any duplicated studies. Two authors (AC-SY) will work independently to extract data from included studies. A standardised, pre-piloted data-extraction form will be used to extract data from included studies. The extracted information will include the author(s), year of study, publication details, study population characteristics (e.g. diagnosis, population size, age, gender, anthropometric measures), FVC, FEV1, MIP, MEP and PCF. Any discrepancies will be discussed, or a third reviewer will be asked to a third reviewer (RTC). Study authors will be contacted with a request for missing data in the included studies.

Subgroup analysis: By age range.

Sensitivity analysis: We will perform sensitivity analysis based on sample size, heterogeneity, methodological quality, and statistical model. We will exclude studies with low quality, and ensure the stability of analysis results.

Language: English.

Country(ies) involved: Chile, Spain.

Keywords: Pulmonary function; Myotonic dystrophy.

Dissemination plans: We will send it to peer review journal.

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