

INPLASY PROTOCOL

To cite: Litwin et al. Maternal and fetal outcomes of pregnancy in Wilson's disease: a systematic review and meta-analysis. Inplasy protocol 202280003. doi: 10.37766/inplasy2022.8.0003

Received: 01 August 2022

Published: 01 August 2022

Corresponding author:
Tomasz Litwin

tomlit@medprakt.pl

Author Affiliation:
Institute Psychiatry and
Neurology, Warsaw, Poland.

Support: None.

Review Stage at time of this submission: Completed but not published.

Conflicts of interest:
None declared.

Maternal and fetal outcomes of pregnancy in Wilson's disease: a systematic review and meta-analysis

Litwin, T¹; Członkowska, A²; Bembenek, J³; Antos, A⁴; Kurkowska-Jastrzębska, I⁵; Przybyłkowski, A⁶; Skowrońska, M⁷; Smolinski, Ł⁸.

Review question / Objective: What are maternal and fetal outcomes of pregnancy in WD?

Condition being studied: The maternal and fetal outcomes of pregnancy in WD: the rates of successful deliveries, abortions, birth defects, and worsening of WD during pregnancy.

Eligibility criteria: We will search the PubMed database (up to 12 January 2022) for original studies (prospective and retrospective), case reports, and case series that have analyzed pregnancy outcomes in women with WD. The search terms will be as follows: ("Wilson's disease"/"Wilson disease" and "pregnancy") and ("Wilson disease"/"Wilson disease" and "birth defect"). Included will be studies published in English. The reference lists of the extracted publications will also be searched.

INPLASY registration number: This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 01 August 2022 and was last updated on 01 August 2022 (registration number INPLASY202280003).

INTRODUCTION

Review question / Objective: What are maternal and fetal outcomes of pregnancy in WD?

Rationale: Several case series, case reports, and retrospective studies have analyzed pregnancy outcomes in WD, including abortion rates, birth defects, and clinical worsening of WD in pregnant women. This systematic review will assess all eligible studies for maternal and fetal

outcomes of pregnancy in WD, what increase the awareness according to WD treatment during pregnancy. The risk of abortion will be compared between treated vs. non-treated patients. The association of birth defects with anti-copper treatment will be investigated.

Condition being studied: The maternal and fetal outcomes of pregnancy in WD: the rates of successful deliveries, abortions, birth defects, and worsening of WD during pregnancy.

METHODS

Search strategy: We will search the PUBMED database for eligible studies.

Participant or population: Pregnant women with WD and their children.

Intervention: We will evaluate the rates of successful deliveries and abortions in patients treated with anti-copper medications and in untreated patients. Additionally, we will relate the rate of birth defects to the use of on anti-copper treatment.

Comparator: Not applicable.

Study designs to be included: Prospective, retrospective studies, case and series reports documenting maternal and fetal outcome of pregnancy in WD.

Eligibility criteria: We will search the PubMed database (up to 12 January 2022) for original studies (prospective and retrospective), case reports, and case series that have analyzed pregnancy outcomes in women with WD. The search terms will be as follows: ("Wilson's disease"/"Wilson disease" and "pregnancy") and ("Wilson disease"/"Wilson disease" and "birth defect"). Included will be studies published in English. The reference lists of the extracted publications will also be searched.

Information sources: Electronic databases.

Main outcome(s): The maternal and fetal outcomes of pregnancy in WD will include: the rates of successful deliveries, abortions, birth defects and worsening of WD during pregnancy. The outcomes will be related to the use of anti-copper treatment.

Quality assessment / Risk of bias analysis: To prevent the risk of bias in this systematic review, a quality assessment tool will be used to assess the quality of all studies included in our analysis.

Strategy of data synthesis: The results and conclusions from all studies included in our analysis will be summarized descriptively or with a random effects meta-analysis.

Subgroup analysis: Treated vs. untreated WD patients.

Sensitivity analysis: In treated vs. untreated patients.

Language restriction: English.

Country(ies) involved: Polska.

Keywords: Wilson's disease, fertility, copper, pregnancy, neurological symptoms, birth defects.

Contributions of each author:

Author 1 - Tomasz Litwin - conception, planning and supervising the work, planning the literature search, selecting the abstracts, extracting data from original papers, writing, revising, approval the manuscript.

Email: tomlit@medprakt.pl

Author 2 - Anna Członkowska - planning the literature search, selecting the abstracts, extracting data from original papers, writing, revising, approval the manuscript.

Email: czlonkow@ipin.edu.pl

Author 3 - Jan Bembenek - planning the literature search, selecting the abstracts, extracting data from original papers, writing, revising, approval the manuscript.

Email: jbembenek@ipin.edu.pl

Author 4 - Agnieszka Antos - planning the literature search, selecting the abstracts,

extracting data from original papers, writing, revising, approval the manuscript.

Author 5 - Iwona Kurkowska-Jastrzębska - planning the literature search, selecting the abstracts, extracting data from original papers, writing, revising, approval the manuscript.

Email: ikurkowska@ipin.edu.pl

Author 6 - Adam Przybyłkowski - planning the literature search, selecting the abstracts, extracting data from original papers, writing, revising, approval the manuscript.

Email: aprzybylkowski@interia.pl

Author 7 - Marta Skowrońska - planning the literature search, selecting the abstracts, extracting data from original papers, writing, revising, approval the manuscript.

Email: mskowronska@ipin.edu.pl

Author 8 - Łukasz Smolinski - planning the literature search, selecting the abstracts, extracting data from original papers, writing, revising, approval the manuscript.

Email: smolinski@ipin.edu.pl