Incidence of Cauda Equina Syndrome: Systematic Review Protocol

ADMINISTRATIVE INFORMATION

Title
Incidence of Cauda Equina Syndrome: Systematic Review Protocol
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Registration
Registered with the International Prospective Register of Systematic Reviews (PROSPERO) on 20th May 2017. Registration Number: CRD42017065865.

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Contributions
• Concept: JW, IH, AKD
• Search strategy: MW, JW, IH
• Reference screening and selection: MW, JW, IH, PC
• Data extraction and quality assessment: MW, JW, IH, PC
• Analysis and report: MW, JW, IH, PC, AKD
• Guarantor: JW, AKD

Amendments
Protocol amendments will be documented, dated, and published in the protocol with the rationale for changes. Amendments will be tracked in PROSPERO.

Support
No external funding will be sought for this systematic review.
INTRODUCTION

Rationale
Cauda equina syndrome (CES) is a clinical syndrome caused by compromise of the cauda equina nerve roots. The syndrome can include bowel, bladder or sexual dysfunction, saddle anaesthesia, and bilateral lower limb sciatica, numbness, or weakness.\(^1\) Operative intervention can prevent symptoms progressing and potentially reverse existing symptoms.\(^2,3\) As the devastating nature of CES leads to significant medical, social, personal and legal costs,\(^4,6\) timely investigation with Magnetic Resonance Imaging (MRI) is recommended to facilitate prompt diagnosis and treatment.\(^7\) However, the clinical symptoms and signs of CES have poor prognostic value, and CES occurs infrequently, which leads to many negative investigations.\(^8,9\) Timely MRI scanning may also require transfer of the patient to tertiary care services, particularly outside normal working hours, which further adds to the resources required to exclude this rare but important syndrome.\(^9\) Establishing the populations in which CES presents and the incidence of radiologically confirmed clinical CES necessitating surgical intervention could aid decision making for the investigation and management of individual patients as well as population service design and delivery.

Objectives
1. To establish the published estimates of the incidence of CES
2. To describe the populations in which the incidence of CES has been studied, and any differences in incidence of CES between these populations
3. To describe the different definitions of CES in use in studies of CES incidence

METHODS

Eligibility Criteria
For inclusion in the review, studies should assess the incidence of CES in humans. All study types reporting original data will be considered. Review articles will not be included unless they also report original data. Clinical CES is defined as any of saddle anaesthesia, bladder dysfunction, bowel dysfunction, sexual dysfunction, or bilateral lower limb symptoms due to the involvement of multiple cauda equina nerve roots. For inclusion as an incident case of CES, patients must have both clinical and radiological CES. Patients with symptoms of CES without identification of a structural cause for these symptoms will not be included as cases. Patients with lesions of the cauda equina without the presence of a clinical CES will not be included as cases. There will be no restrictions on the type, location, or age of the populations included. There will be no language or date restrictions on studies. Both published studies and unpublished studies will be considered.

Information Sources
The search will be carried out in MEDLINE (Ovid), EMBASE (Ovid), and Scopus. The reference lists of all included studies will be hand searched to identify any other relevant papers. Papers that have cited any of the included studies will be identified in Scopus and any relevant papers included.
Search Strategy
Preliminary searches were used to identify relevant subject headings and refine the search strategy with the help of a medical librarian. Proposed draft search strategies for each database are shown below. No limits will be applied to the searches. Searches will be updated at the end of the review process to ensure any recently published material is included.

MEDLINE
1. Polyradiculopathy/
2. cauda equina.ti,ab.
3. Cauda Equina/
4. 1 OR 2 OR 3
5. Incidence/ or Prevalence/
6. Epidemiology/
7. (incidence* or prevalen* or epidemiolog* or frequenc* or rate* or occurrence*).ti,ab
8. 5 OR 6 OR 7
9. 4 AND 8

EMBASE
1. Cauda equina syndrome/
2. Cauda equina.ti,ab
3. Cauda Equina/
4. 1 OR 2 OR 3
5. Incidence/ or Prevalence/
6. Epidemiology/
7. (incidence* or prevalen* or epidemiolog* or frequenc* or rate* or occurrence*).ti,ab
8. 5 OR 6 OR 7
9. 4 AND 8

SCOPUS
1. “cauda equina“
2. (incidence* or prevalen* or epidemiolog* or frequenc* or rate* or occurrence*)
3. 1 AND 2

Study Records
References identified through the search will be imported into Endnote. Duplicates will be identified and eliminated in Endnote. Abstracts and titles will be screened independently by at least two review team members. Any disagreements will be solved by discussion with a third review team member. The full text will be sourced for all abstracts that are identified as being potentially relevant, or where inclusion or exclusion cannot be determined from the abstract. Full text articles will be screened independently by two review team members. Any disagreements on inclusion or exclusion will be solved through discussion. The reason for exclusion of full text articles will be recorded. Data will be extracted from each article using a standardised form. Two review team members will extract data independently, and any discrepancies will be checked by a third review team member. We will contact study authors to resolve any uncertainties in the data. Where there is more than one record of a single study, that study will only be entered once into the analysis, but multiple records may be used for data extraction.
Data Items
Data that will be extracted from each included study will include:

- incidence of CES in the population studied
- confidence intervals of incidence figures
- standardised estimates of incidence figures
- number of cases of radiologically confirmed clinical CES
- definition of CES used in the study
- demographics of patients with CES
- size of the reference population
- description of the reference population
  - country or location
  - demographics
  - inclusion criteria for reference population
  - methods of establishing the reference population
  - reasons for any exclusions from the reference population
  - description of subjects excluded from the reference population
  - time period over which population was studied

Outcomes and Prioritisation
The primary outcome is the estimate of the incidence of CES. Secondary outcomes are the description of the populations in which the incidence of CES has been studied, the different incidence figures for CES within these populations, and the different definitions of CES in use. Where more than one report of a single study is available, the following priorities will be used to select the data for inclusion: the most complete version with the largest population; the version giving the data most relevant to the inclusion criteria; published data.

Risk of Bias in Individual Studies
Each study will be assessed for quality and risk of bias using a set of questions adapted from those used in prior systematic reviews of incidence of neurological conditions,\textsuperscript{10, 11} and based on published quality assessment guidelines.\textsuperscript{12, 13} The following questions will be answered for each study.

1. Was the target population clearly described?
2. Were cases ascertained by survey of the entire population or by probability sampling?
3. Was the sample size >300 subjects?
4. Was the response rate >70%?
5. Were non-responders clearly described?
6. Was the sample representative of the population?
7. Were data collection methods standardised?
8. Were the diagnostic criteria used to assess the presence / absence of disease described?
9. Were estimates of incidence given with confidence intervals?
10. Were standardised estimates reported?
As there are no validated diagnostic criteria for the presence of clinical CES, question seven has been changed from asking whether validated diagnostic criteria were used to an assessment of whether the definition of clinical CES used in the study was described.

Data Synthesis
Initially, a systematic narrative description of included studies will be given, with incidence figures presented in text and tables. The populations in which the incidence of CES has been studied will be identified, and incidence figures for CES presented separately for these populations. Incidence figures will be analysed and compared within similar populations. If a sufficient number of studies of similar populations are identified, and these studies report standard errors of incidence figures, then meta-analysis will be undertaken. Heterogeneity of studies will be analysed using the Q statistic and the I² test.

Meta-Bias
Publication bias will be assessed using visual inspection of funnel plots.

Confidence in Cumulative Evidence
The quality of evidence for the incidence figures will be assessed using the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) approach.¹⁴

REFERENCES


