What is the incidence of cauda equina syndrome? A systematic review

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OBJECTIVE Cauda equina syndrome (CES) is a surgical emergency requiring timely operative intervention to prevent symptom progression. Accurately establishing the incidence of CES is required to inform healthcare service design and delivery, including out-of-hours imaging arrangements.

METHODS A systematic literature search of MEDLINE, EMBASE, and Scopus was undertaken to identify original studies stating the incidence of CES, and the estimates were combined in a meta-analysis as described in the protocol registered with PROSPERO (registration no. CRD42017065865) and reported using the PRISMA guidelines.

RESULTS A total of 1281 studies were identified, and 26 studies were included in the review. Data about CES incidence were available from 3 different populations: asymptomatic community populations, patients with nontraumatic low-back pain, and patients presenting as an emergency with suspected CES. The incidence of CES was 0.3–0.5 per 100,000 per year in 2 asymptomatic community populations, 0.6 per 100,000 per year in an asymptomatic adult population, and 7 per 100,000 per year in an asymptomatic working-age population. CES occurred in 0.08% of those with low-back pain presenting to primary care in 1 study, and a combined estimate of 0.27% was calculated for 4 studies of those with low-back pain presenting to secondary care. Across 18 studies of adults with suspected CES, 19% had radiological and clinical CES. Difficulties in comparison between studies resulted from the heterogeneous definitions of CES and lack of separation of more advanced CES with retention, which is unlikely to be reversible. In the studies of patients with suspected CES, the small sample size, the high number of single-center studies (18/18), the high number of studies from the United Kingdom (17/18), the retrospective nature of the studies, and the high number of abstracts rather than full texts (9/18) reduced the quality of the data.

CONCLUSIONS From current studies, it appears that CES occurs infrequently in asymptomatic community populations and in only 19% of those presenting with symptoms. Determining accurate incidence figures and designing a bespoke service for investigation of patients with suspected CES would require a consensus clinical and radiological definition of CES and international multisite studies of patient pathways of investigation and management.


KEYWORDS cauda equina syndrome; incidence; systematic review; epidemiology; population; lumbar

CAUDA equina syndrome (CES) is a surgical emergency with potentially significant consequences, including bladder, bowel and sexual dysfunction, numbness, weakness, and pain. Timely operative intervention can prevent symptom progression and potentially reverse existing symptoms. Due to the high medical, personal, social, and legal costs, prompt investigation with MRI is recommended when CES is suspected. In the United Kingdom (UK), patients are often transferred for investigation between sites due to a lack of MRI facilities operating outside normal working hours in district general hospitals and the potential need for specialist spinal or neurosurgical intervention. However, many patients who present with clinical symptoms in keeping with CES will not have cauda equina compression on MRI, which complicates planning service design and delivery. Establishing the incidence of CES and the likelihood of a diagnosis of CES in those presenting with symptoms consistent with CES would facilitate planning imaging and operative pathways for patients with suspected CES.
In this systematic review, we aimed to identify studies reporting the incidence of CES and describe the populations in which the incidence of CES has been studied and any differences in incidence between these populations.

**Methods**

A systematic review was undertaken as described in the study protocol “Incidence of cauda equina syndrome: systematic review protocol” registered with the International Prospective Register of Systematic Reviews (PROSPERO; reference no. CRD42017065865, available at https://www.crd.york.ac.uk/PROSPERO/display_record.php?RecordID=65865).

Studies were included if they reported original data and assessed human subjects with CES. For inclusion, studies had to state the incidence of CES or the proportion of the studied population with CES, or provide sufficient figures for this to be calculated. We defined CES as a clinical diagnosis of CES with radiological cauda equina compression. Studies including only patients with a clinical CES-type syndrome without radiologically confirmed cauda equina compression were excluded. Studies of radiological lesions of the cauda equina or cauda equina compression without clinical features of CES were also excluded. Reference populations could be either asymptomatic or symptomatic populations. Case series or studies without a reference population where the incidence of CES could not be established were excluded. Case series of operated lumbar discs, spinal stenosis, or iatrogenically caused CES were also excluded to ensure that all included studies were applicable to an initial presentation with suspected CES. There were no restrictions on the language or year of publication; type, location, or age of the population studied; or whether the study was published or unpublished.

The final database search was carried out on July 30, 2018, in MEDLINE (Epub Ahead of Print, In-Process & Other Non-Indexed Citations, and Daily 1946 to July 27, 2018, Ovid interface); EMBASE (1980 to week 31 of 2018, Ovid interface); and Scopus. The MEDLINE search strategy was as follows:

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 prevalence* or epidemiology* or frequency* or rate* or occurrence*),ti,ab
8. 5 OR 6 OR 7
9. 4 AND 8

No limits were applied. EMBASE and Scopus search strategies are given in the Appendix.

Duplicate studies were eliminated, and then all abstracts and titles were screened by 2 reviewers independently (J.W., I.H., P.C.C., or M.W.). When reviewers disagreed, discussion with a third or fourth reviewer was undertaken to provide a consensus. The full text of all included abstracts was retrieved and independently reviewed by 2 reviewers (J.W., I.H., P.C.C., or M.W.). Any disagreements were resolved through discussion with a third or fourth reviewer. The reference lists of all included studies were screened independently by 2 reviewers to identify any additional relevant papers. Studies citing the included studies were identified using Scopus and also screened by 2 reviewers independently. Multiple papers or abstracts reporting the same study were treated as a single study.

Data were extracted from each included paper by 2 reviewers independently, and all instances in which data did not match were checked by a third reviewer (J.W., I.H., P.C.C., or M.W.). The data items extracted were incidence of CES in the population (including confidence intervals and standardized estimates where given); number of cases of CES; size of the reference population; description of the population (location, demographics, time period studied, inclusion criteria); and definition of CES used in the study including any subcategorization.

Study quality and risk of bias were assessed using the following questions adapted from those used in prior systematic reviews of the incidence of neurological conditions based on published quality assessment guidelines. As there are no validated diagnostic criteria for CES, studies were assessed on whether they described the definition of CES used.

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 nonresponders clearly described?
6. Was the sample representative of the population?
7. Were data collection methods standardized?
8. Were the diagnostic criteria used to assess the presence of CES defined?
9. Were estimates of incidence given with confidence intervals?
10. Were standardized estimates reported?

The incidence of CES was reported per 100,000 population per year in asymptomatic populations. The percentage of CES was reported in symptomatic populations. Statistical heterogeneity was assessed using the Q statistic and the I² test. Proportions were combined using the inverse variance method and a DerSimonian-Laird estimator for τ². Confidence intervals for individual studies were calculated using Clopper-Pearson confidence intervals. All statistics were calculated using the meta package in R version 3.4.0.

**Results**

The studies identified and excluded at each stage and reasons for exclusion are shown in the PRISMA flow diagram (Fig. 1). Of the 1281 studies identified after removal of duplicates, 26 were included, of which 21 were included in the meta-analysis. Four studies reported the incidence of CES occurring in asymptomatic community populations. Twenty-three studies investigated the incidence of CES in patients presenting with symptoms. One study was included in both of these categories.
Population Incidence of CES

Study details and incidence figures for the 4 studies reporting the incidence of CES in community-dwelling asymptomatic populations are shown in Table 1. Hurme et al.\textsuperscript{28} and Podnar\textsuperscript{37} investigated European community-dwelling populations and identified similar incidence figures of 0.48 and 0.34 cases per 100,000 population per year, respectively, despite different methods of case ascertainment. Hurme et al.\textsuperscript{28} identified cases of CES using surgical records, while Podnar\textsuperscript{37} used a comprehensive clinical and neurophysiological assessment at a rehabilitation center. Reito et al.\textsuperscript{40} reported the incidence in an adult-only population and found a slightly higher incidence of 0.6 per 100,000 adult population per year. Schoenfeld\textsuperscript{42} and Schoenfeld and Bader\textsuperscript{43} studied an American military personnel healthcare database and found a higher incidence of 7 per 100,000 population per year in this working-age population. The study by Reito et al.\textsuperscript{40} was the only one to divide CES into subcategories. Two patients had CES with retention and 2 patients had incomplete CES, making the incidence of each subtype 0.30 per 100,000 per adult population per year. Both Reito et al. and Schoenfeld and Bader\textsuperscript{43} used coding to identify cases of CES. Reito et al. also reviewed clinical notes of the identified cases. Meta-analysis of the incidence estimates was not undertaken due to the heterogeneity in the reference populations studied and the methods of CES case ascertainment.

Incidence of CES in Patients With Back Pain

Five studies reported the proportion of patients presenting with nontraumatic low-back pain who were found to have CES.\textsuperscript{23,30,38,40,48} Study findings are shown in Table 2. Henschke et al.\textsuperscript{23} found that 0.08% of adults presenting to primary care in Australia with low-back pain were...
diagnosed with CES by the study rheumatologist using clinical assessment and investigation. The other 4 studies investigated patients presenting to secondary care and reported proportions between 0.15% and 0.54%. The diagnosis of CES was determined by ICD code in 2 studies and by the clinician in 1 study; the method was not reported in 1 study. Study estimates for the proportion with CES in those presenting to secondary care with nontraumatic low-back pain were combined using a random-effects model to give an estimated proportion of 0.27% (95% CI 0.14%–0.54%). Study estimates and confidence intervals are shown in the forest plot in Fig. 2. There was a high level of statistical heterogeneity, with $I^2 = 85.2\%$ (95% CI 63.3%–94.0%) and $Q = 20.2$ ($p < 0.001$).

### Incidence of Confirmed CES in Patients Suspected of Having CES

Eighteen studies reported the proportion of patients presenting with signs and symptoms suspicious for CES who had clinical and radiological confirmation of CES. The study details are shown in Table 3. Ten studies included only patients undergoing MRI for suspected CES. Two studies included patients undergoing urgent spinal MRI, but did not specify whether this was to investigate CES in every case. Six studies included all patients referred with suspected CES. All studies assessed populations referred to either secondary or tertiary care. Banerjee and Jalgaonkar studied only children. All other studies included adult populations but did not state whether they specifically excluded pediatric patients. A diagnosis of CES was established by cauda equina compression on MRI or operative intervention for CES. The imaging type in all studies was MRI. Only 2 studies described findings on MRI defining a diagnosis of CES based on the finding of canal compromise, which was more than 50% in one study and more than 75% in the other. Three studies stated that cauda equina compression was determined by the reporting radiologist but did not state the criteria used. Causes of cauda equina compression were described in 6 studies. Demetriades et al. only included disc prolapses. Five studies included all or some of disc prolapses, tumors, trauma, and hematoma. One study discussed subtypes of CES (with urinary symptoms or incomplete CES) but did not report the numbers in each group. None of the other studies used subcategories or descriptors. Four studies provided information on symptom duration. Urinary symptoms in 2 studies were present for an average of 4 and 5.8 days, and symptoms not further specified were present for between 24 hours and 6 months and a median.

### TABLE 1. Incidence of CES in asymptomatic community populations

<table>
<thead>
<tr>
<th>Authors &amp; Study</th>
<th>Yrs of Study</th>
<th>Time Period</th>
<th>Reference Population</th>
<th>Definition of CES</th>
<th>Total Population</th>
<th>Total Cases</th>
<th>Cases/100,000/Yr (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hurme et al., 1983</td>
<td>1975–1979</td>
<td>5 yrs</td>
<td>Hospital catchment population, Finland</td>
<td>Undergoing operation for CES</td>
<td>455,000</td>
<td>11</td>
<td>0.48*</td>
</tr>
<tr>
<td>Podnar, 2007</td>
<td>1996–2004</td>
<td>8 yrs</td>
<td>Population of Slovenia</td>
<td>History, examination, neurophysiology &amp; radiology</td>
<td>1,989,198</td>
<td>67</td>
<td>0.34</td>
</tr>
<tr>
<td>Reito et al., 2018</td>
<td>2012–2014</td>
<td>3 yrs</td>
<td>Hospital catchment population, Finland</td>
<td>ICD code; SBNS guideline subcategories based on clinical records</td>
<td>661,902 adult person-yrs†</td>
<td>4</td>
<td>0.6 (0.16–1.5)</td>
</tr>
</tbody>
</table>

SBNS = Society of British Neurological Surgeons.

* Calculated from values given in paper.
† Reported as the total number of people in the population in the total number of years during the study time period.

### TABLE 2. Incidence of CES in patients presenting with back pain

<table>
<thead>
<tr>
<th>Authors &amp; Study</th>
<th>Yrs of Study</th>
<th>Time Period</th>
<th>Reference Population</th>
<th>Definition of CES</th>
<th>Total Population</th>
<th>Total Cases</th>
<th>Proportion w/ CES (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Henschke et al., 2009</td>
<td>2003–2005</td>
<td>20 mos</td>
<td>Primary care, Australia</td>
<td>Rheumatologist assessment (history, exam, tests)</td>
<td>1172</td>
<td>1</td>
<td>0.08% (0.0–0.5%)</td>
</tr>
<tr>
<td>Thiruganasambandamoorthy et al., 2014</td>
<td>2009–2010</td>
<td>3 mos</td>
<td>Adults, ED, Canada</td>
<td>Clinician determined</td>
<td>329</td>
<td>1</td>
<td>0.30%</td>
</tr>
<tr>
<td>Kiberd et al., 2018</td>
<td>Not stated</td>
<td>7 yrs</td>
<td>ED, Canada</td>
<td>Not stated</td>
<td>38,714</td>
<td>57</td>
<td>0.15%</td>
</tr>
<tr>
<td>Premkumar et al., 2018</td>
<td>2005–2016</td>
<td>11 yrs</td>
<td>Spinal surgeon, US</td>
<td>ICD code</td>
<td>9940</td>
<td>36</td>
<td>0.36%</td>
</tr>
<tr>
<td>Reito et al., 2018</td>
<td>2012–2014</td>
<td>3 yrs</td>
<td>Adults, ED, Finland</td>
<td>ICD code; SBNS guideline subcategories – based on clinical records</td>
<td>900 visits; 737 patients</td>
<td>4</td>
<td>0.44% per visit; 0.54% per patient</td>
</tr>
</tbody>
</table>

ED = emergency department.
**FIG. 2.** Forest plot. Proportion and number (events) of patients with CES among those presenting with nontraumatic low-back pain to secondary care. Summary proportion calculated using a random effects model.

**TABLE 3. Incidence of CES in patients presenting with suspected CES**

<table>
<thead>
<tr>
<th>Authors &amp; Study</th>
<th>Yrs of Study</th>
<th>Time Period</th>
<th>Reference Population: Potential CES</th>
<th>Definition of CES</th>
<th>Imaging Type</th>
<th>Total Population</th>
<th>Total Cases</th>
<th>Proportion w/ CES</th>
<th>Weight</th>
</tr>
</thead>
<tbody>
<tr>
<td>Thiruganasambandamoorthy, 2014</td>
<td>Not stated</td>
<td>4 mos</td>
<td>MRI for ?CES, neurosurgery, UK</td>
<td>MRI CE compression</td>
<td>MRI</td>
<td>23</td>
<td>5</td>
<td>21.7%</td>
<td></td>
</tr>
<tr>
<td>Kiberd 2018</td>
<td>Not stated</td>
<td>2 yrs</td>
<td>OOH MRI for ?CES, neurosurgery, UK</td>
<td>Surgery for CES</td>
<td>MRI</td>
<td>82</td>
<td>27</td>
<td>32.9%</td>
<td></td>
</tr>
<tr>
<td>Premkumar 2018</td>
<td>Not stated</td>
<td>1 yr</td>
<td>OOH MRI for ?CES, neurosurgery, UK</td>
<td>Disc on MRI &amp; surgery for CES</td>
<td>MRI</td>
<td>33</td>
<td>10</td>
<td>30.3%</td>
<td></td>
</tr>
<tr>
<td>Reito 2018</td>
<td>Not stated</td>
<td>5 yrs</td>
<td>Urgent MRI for ?CES neurology/ED, the Netherlands</td>
<td>Radiology report MRI CE compression</td>
<td>MRI</td>
<td>58</td>
<td>8</td>
<td>13.8%</td>
<td></td>
</tr>
<tr>
<td>Rooney et al., 2009</td>
<td>Not stated</td>
<td>10 mos</td>
<td>MRI for ?CES, neurosurgery, UK</td>
<td>Surgery for CES</td>
<td>MRI</td>
<td>66</td>
<td>16</td>
<td>24.2%</td>
<td></td>
</tr>
<tr>
<td>Balasubramanian et al., 2008</td>
<td>2008</td>
<td>1 yr</td>
<td>MRI for ?CES, spinal surgery, UK</td>
<td>Radiology report &gt;75% canal compromise</td>
<td>MRI</td>
<td>80</td>
<td>15</td>
<td>18.5%</td>
<td></td>
</tr>
<tr>
<td>Thangarajah et al., 2011</td>
<td>2008–2007</td>
<td>1 yr</td>
<td>Urgent spinal MRI, teaching hospital, UK</td>
<td>Radiology report CE compression</td>
<td>MRI</td>
<td>81</td>
<td>0</td>
<td>0%</td>
<td></td>
</tr>
<tr>
<td>Gooding et al., 2013</td>
<td>Not stated</td>
<td>1 yr</td>
<td>MRI for ?CES, hospital w/ spinal unit, UK</td>
<td>Radiology report CE compression</td>
<td>MRI</td>
<td>57</td>
<td>13</td>
<td>22.8%</td>
<td></td>
</tr>
<tr>
<td>Haworth et al., 2009</td>
<td>2009–2011</td>
<td>3 yrs</td>
<td>OOH MRI for ?CES, neurosurgery, UK</td>
<td>MRI CE compression</td>
<td>MRI</td>
<td>162</td>
<td>39</td>
<td>24.1%</td>
<td></td>
</tr>
<tr>
<td>Sideris et al., 2014</td>
<td>2010–2013</td>
<td>4 yrs</td>
<td>?CES, neurosurgery, UK</td>
<td>Clinical &amp; radiological CES</td>
<td>MRI</td>
<td>663</td>
<td>80*</td>
<td>12.0%</td>
<td></td>
</tr>
<tr>
<td>Ahad et al., 2015</td>
<td>2012–2013</td>
<td>8 mos</td>
<td>Urgent spinal MRI, hospital, UK</td>
<td>MRI CE compression</td>
<td>MRI</td>
<td>79</td>
<td>5</td>
<td>6.3%</td>
<td></td>
</tr>
<tr>
<td>Blades et al., 2015</td>
<td>2008–2014</td>
<td>7 yrs</td>
<td>?CES, spinal unit, UK</td>
<td>MRI CE compression</td>
<td>MRI</td>
<td>344</td>
<td>137</td>
<td>40%</td>
<td></td>
</tr>
<tr>
<td>Hoeritzauer et al., 2015</td>
<td>2013–2014</td>
<td>6 mos</td>
<td>Urgent MRI for ?CES spinal unit, UK</td>
<td>MRI CE compression</td>
<td>MRI</td>
<td>18</td>
<td>7</td>
<td>38.9%</td>
<td></td>
</tr>
<tr>
<td>Hoeritzauer et al., 2017</td>
<td>2013–2014</td>
<td>16 mos</td>
<td>?CES, neurosurgery, UK</td>
<td>MRI CE compression</td>
<td>MRI</td>
<td>290</td>
<td>91</td>
<td>31.4%</td>
<td></td>
</tr>
<tr>
<td>Kostusiak et al., 2018</td>
<td>2014–2017</td>
<td>4 yrs</td>
<td>OOH MRI for ?CES, neurosurgery, UK</td>
<td>Radiology report CE compression</td>
<td>MRI</td>
<td>323</td>
<td>15</td>
<td>4.6%</td>
<td></td>
</tr>
<tr>
<td>Hussain et al., 2018</td>
<td>2013–2014</td>
<td>14 mos</td>
<td>?CES, neurosurgery, UK</td>
<td>&gt;50% canal compromise on MRI</td>
<td>MRI</td>
<td>250</td>
<td>32</td>
<td>12.8%</td>
<td></td>
</tr>
<tr>
<td>Banerjee, 2018</td>
<td>2014–2016</td>
<td>3 yrs</td>
<td>?CES, district hospital, UK</td>
<td>MRI CE compression</td>
<td>MRI</td>
<td>43</td>
<td>7</td>
<td>16.3%</td>
<td></td>
</tr>
<tr>
<td>Banerjee &amp; Jalgaonkar, 2018</td>
<td>2012–2017</td>
<td>5 yrs</td>
<td>Children (0–15 yrs), ?CES, district hospital, UK</td>
<td>MRI CE compression</td>
<td>MRI</td>
<td>15</td>
<td>0</td>
<td>0%</td>
<td></td>
</tr>
</tbody>
</table>

*CES = potential CES; CE = cauda equina; OOH = out of hours.
* Calculated from paper.
of 11 days in 2 other papers. Two studies containing small numbers of patients with CES investigated whether any symptoms or signs were predictive of CES. In the 6 patients assessed with bladder scanning, Domen et al. found that urinary retention of > 500 ml plus at least 2 of bilateral sciatica, subjective urinary retention, or rectal incontinence had an odds ratio of 48 for predicting cauda equina compression on MRI.15 In 5 patients with CES, Ahad et al. did not find any predictive symptoms but found that patients with abnormal MRI findings of the spine for back pain prior to CES presentation were significantly more likely to have radiological compression.1 These results are limited in their generalizability by the small numbers of patients involved. The proportion with confirmed CES in those presenting with suspected CES ranged from 0% to 40% in the 18 studies. We excluded the study that included only children5 and combined the other estimates using a random-effects model to give an overall estimate of confirmed CES in 18.9% (95% CI 13.6%–25.6%). The forest plot is shown in Fig. 3. There was a high level of heterogeneity in the study designs and the statistical heterogeneity was high, with $I^2 = 91.9\%$ (95% CI 88.6%–94.3%) and $Q = 197$ ($p < 0.001$).

### Study Quality

Study quality assessment is shown in Table 4. All studies described the population being studied and had representative samples. However, definitions of CES and methods used to ascertain the diagnosis of CES varied between studies, and many studies did not adequately describe their methods in a way that could be easily reproduced. Only 2 studies reported excluded patients,40,41 and only 1 study described the excluded patients.40 Only 2 studies calculated confidence intervals for the incidence estimates, 23,40 and none reported population-standardized estimates. Of the 26 studies included in this review, 9 were published only in abstract form.4,5,7,13,22,27,30,32,45 Studies of patients with suspected CES were of particularly poor quality. They were limited by small sample sizes; only 3 (17%) studies included more than 300 participants,7,32,45 by their retrospective (100%) and single-center design (100%), and by the limited information available, as so many (50%) were published only as an abstract. Of the 18 studies of patients with suspected CES, all but one were from the UK.

### Discussion

This is the first systematic review of studies estimating the incidence of CES; 26 studies were included. The incidence of CES is low, at fewer than 1 per 100,000 people in asymptomatic populations per year. Only 0.27% of those with low-back pain and only 18.9% of those with signs and symptoms consistent with CES will have a final diagnosis of radiological and clinical CES.

This review identified a paucity of literature on the incidence of CES. We included all studies from which inci-
## TABLE 4. Study quality and risk of bias in included studies

<table>
<thead>
<tr>
<th>Authors &amp; Study</th>
<th>Target Population</th>
<th>Cases From Entire Population</th>
<th>Sample Size &gt;300</th>
<th>Response Rate &gt;70%</th>
<th>Nonresponders Clearly Described</th>
<th>Sample Representative</th>
<th>Standardized Data Collection</th>
<th>Diagnostic Criteria Described</th>
<th>Estimates Given w/ Confidence Intervals</th>
<th>Standardized Estimates Reported</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hurme et al., 1983</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>?</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>Podnar, 2007</td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
<td>?</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>Schoenfeld &amp; Bader, 2012</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>?</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>Reito et al., 2018</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>Henschke et al., 2009</td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
<td>?</td>
<td>No</td>
<td>Yes</td>
<td>Yes</td>
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? = no information given.

Studies were assessed against the 11 prespecified criteria.
idence of CES could be calculated, but few of the studies had a primary aim to calculate incidence. Many did not meet expected epidemiological standards, as can be seen from Table 4. Sample sizes were small in symptomatic populations and estimates did not have confidence intervals and were not standardized for the populations. Few studies described exclusions or missing data. Nine studies were only published in abstract form and provided fewer methodological details and had not been through the peer-review process. All abstracts and full-text articles were screened by at least 2 reviewers, and we only identified 7 additional studies through searching reference lists and citations. We are confident that these methods should not have missed any further important studies on this topic.

The criteria used to establish a diagnosis of CES were described in only 13 of the 26 studies, and only 2 studies subdivided CES into clinical categories. Diagnosis was determined through clinical coding, record review, urgent operative intervention, radiology reports, clinical assessment, or any combination of these. The variation in definitions and reporting of diagnostic criteria likely reflects the lack of agreed definitions and multiple classifications of CES in use clinically and in the literature. The lack of specific clinical phenotyping covered by a broad CES definition hampers accurate assessment of incidence and contributes to the statistical heterogeneity as the incidence will likely differ depending on the definition and case ascertainment methods used. Adopting agreed-on definitions or defining subtypes such as those listed by Todd and Dickson might enable more consistent reporting in future studies and allow more accurate incidence figures to be established.

One study was carried out in Australia, and 4 studies were carried out in North America, and the remaining studies were in European populations. It is not known whether these estimates are relevant outside the populations and healthcare settings studied. Location may determine the availability of imaging and clinical threshold for investigation. All but one study reporting the proportion of patients with CES from those with suspected CES were carried out in the UK. This may reflect the interest in determining the yield of MRI scanning for suspected CES in a healthcare setting where access to out-of-hours MRI is not always readily available. Guidance from the British Association of Spine Surgeons recommends an emergency MRI for suspected CES, and yet only 14% of hospitals in England and Wales surveyed in 2012 reported 24-hour access to MRI. As clinical symptoms and signs in those with radiological cauda equina compression are very difficult to distinguish from those without cauda equina compression, this leads to a situation in which many patients are transferred to specialist centers for MRI outside office hours and then either transferred back or discharged from locations that can be far from home. In healthcare settings with pressure on MRI services, such as the UK, the threshold for investigating patients with MRI for suspected CES may be higher than in a situation in which MRI is quickly and readily available 24 hours a day. It is not known whether easy access to MRI correlates with a lower diagnostic yield of positive scans for cauda equina compression on MRI due to an increased overall number of patients undergoing MRI.

Healthcare service planning for the investigation and management of CES needs to balance the needs of the majority population with the few CES cases in whom a missed diagnosis or delayed treatment could have significant health and social care consequences for the patient plus medicolegal consequences for the surgeon and healthcare service. Different medicolegal implications in different countries may affect the threshold for investigating and diagnosing CES, which will ultimately affect estimates of incidence. Between 2013 and 2016, there were 131 claims relating to CES in the UK, with a projected value of $68 million. These were most commonly due to delay in diagnosis or treatment. In the US, the average payout of 15 lawsuits related to CES between 1983 and 2010 was $1.57 million. It is unknown whether the frequency of legal action for CES in a country is associated with the clinical threshold for investigation of symptoms with an MRI study, as all but one study of patients with suspected CES were carried out in the UK. The high legal costs to the health service of a missed case must be weighed against the costs involved in implementing systems to ensure timely MRI in patients with suspected CES.

Most patients investigated for suspected CES do not have radiological compression on MRI. Although final diagnoses in patients without cauda equina compression include demyelination, myelitis, and infection, a structural cause is not found in the majority of patients. Further characterization of these patients to identify potentially distinguishing features such as Hoover’s sign of functional weakness could increase the yield of MRI for suspected CES. However, due to the significance of a missed diagnosis, expansion of local out-of-hours MRI provision is more likely to improve care for those investigated for CES with and without structural radiological cauda equina compression. Local MRI services would avoid unnecessary transfer of patients to tertiary services that they do not require.

Conclusions

CES occurs infrequently in asymptomatic communities and in only 19% of those presenting with symptoms. Major limitations in the published literature make it difficult to provide evidence-based services for patients with CES. Multicenter and international studies are required. However, before these can occur, a consensus definition of CES, including clinical and radiological criteria, is needed to allow comparison across centers and throughout the literature.

Acknowledgments

We thank Sheila Fisken, University of Edinburgh librarian, for her support in designing and developing the search strategy.

Appendix

Search Strategies

Ovid EMBASE: 1980 to 2018 week 31
1) Cauda equina syndrome/
2) cauda equina.ti,ab.
3) Cauda Equina/
References


Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: Woodfield, Hoeritzauer, Demetriades. Acquisition of data: Woodfield, Hoeritzauer, Wood, Copley. Analysis and interpretation of data: Woodfield, Hoeritzauer, Wood, Copley. Drafting the article: Woodfield, Hoeritzauer, Wood, Copley. Critically revising the article: Woodfield, Hoeritzauer, Demetriades. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Woodfield. Statistical analysis: Woodfield, Wood. Administrative/technical/material support: Hoeritzauer, Wood. Study supervision: Woodfield, Hoeritzauer, Demetriades.

Supplemental Information
Previous Presentations
Portions of this paper were presented in poster form at the Society of British Neurological Surgeons, April 11, 2018, Plymouth, United Kingdom.

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