

# A multi-perspective approach to cauda equina syndrome dedicated to sex, micturition and defecation

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# **CHAPTER 2**

# Complaints of micturition, defecation and sexual function in cauda equina syndrome due to lumbar disk herniation: a systematic review

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# ABSTRACT

*Purpose*. Cauda equina syndrome (CES) is a rare complication of lumbar disc herniation. Although micturition, defecation and/or sexual function are by definition affected, little is known about long term outcome. Aim of this study is to review current literature on outcome of micturition, defecation and sexual function in CES due to lumbar disc herniation.

*Methods.* A literature search was done in Pubmed, Embase and Web of Science using a sensitive search string combination. Studies were selected by predefined selection criteria and risk of bias was assessed using a Cochrane checklist adjusted for this purpose. *Results.* Fifteen studies were included. Risk of bias varied with six studies showing low risk. Mean minimal follow up time was 17.0 months (range 3-25 months). All studies evaluated micturition and reported dysfunction at follow up in 42.5% (range 13.3-90.0%). Defecation and sexual function were evaluated in eight and nine studies, respectively, and were reported to be 49.6% (range 10.5-90.0%) and 44.3% (range 10.0-76.6), respectively. Only two studies assessed sexual function in all patients at follow up.

*Conclusion.* This review offers an insight into the extent of micturition dysfunction, defecation dysfunction and sexual dysfunction in CES after decompressive surgery. Our findings show that dysfunction is extremely common, even at long term follow up. A condition as invalidating as CES requires proper patient information and the outcomes presented here may help in providing those data. Bias in included studies, lack of universal definitions and incomplete follow up results qualify these data as the best we momentarily have, but still subject to improvement. Since sexual dysfunction (SD) seems to be severely underreported, we recommend further research to explore the extent of this problem, as well as the use of questionnaires in future clinical (prospective) studies to accomplish a more patient-based approach to dysfunction.

#### INTRODUCTION

Cauda equina syndrome (CES) is an uncommon neurologic condition caused by compression of the cauda equina. The first notion of CES in English literature was taken in 1934 by Mixter and Barr.<sup>1</sup> They described a spectrum of neurological and urological complaints in patients with a lumbar herniated disc and attributed this to a severe compression of the cauda equina, urging for emergency decompression as opposed to a more expectative treatment in uncomplicated hernia nuclei pulposi (HNP). Although CES can be instigated by any pathological process compressing the cauda equina, the lumbar disc is the most prevalent structure: 45% of described CES cases in literature are attributed to the disc.<sup>2</sup> In patients operated on because of HNP, the incidence of CES is 1-10%.<sup>3,4</sup> Because CES is an indication for surgery in HNP, the incidence of CES among general HNP patients is probably lower.<sup>5</sup>

Exact definitions of CES have always been a topic of ambiguity, although most authors agree that micturition dysfunction should be present.<sup>6-9</sup> After reviewing more than a hundred articles, Fraser et al. stated that at least one or more of the following should be present to diagnose CES: (1) micturition and/or defecation dysfunction, (2) reduced sensation in the saddle area, and (3) sexual dysfunction (SD) with possible neurologic deficit in the lower limb (motor/sensory loss, reflex change).<sup>2</sup> Even though the relevance of micturition dysfunction, defecation dysfunction and sexual dysfunction is clear from this definition, little seems to be known about the long term outcome of these functions. This poses a problem for the clinician who needs to provide his or her patient with an accurate prognosis and thereby obtain true informed consent for therapy.

Solid information about long term recovery of micturition, defecation and sexual function in CES is not abundant. One factor is probably the shame that revolves around discussing problems of micturition, defecation and sexual function. Secondly, the extensive focus in literature on the timing of decompression, pushes the actual hard figures on long term outcome more to the background. For the surgeon confronted with a CES patient, however, questions do not arise around the best timing of surgery, since it is generally accepted that decompression should be done as soon as possible. Questions do arise around the long term prognosis. More specifically, the prognoses of micturition, defecation and sexual function, which are so inevitably affected in CES, need to be clarified. Individual studies at best provide the clinician with uncertain estimates based on few patients. The aim of this study is therefore to review current literature on the outcome of micturition, defecation and sexual function and sexual function in CES due to HNP.

# **MATERIAL AND METHODS**

### Data searches and study selection

In January 2012, the electronic databases Pubmed, Embase and Web of Science were searched using the search strategies as shown in Figure 1.

| PUBMED (149 results)<br>("lumbar disk herniation" OR "lumbar disc herniation" OR "lumbar herniated disk" OR "lumbar herniated disc" OR<br>"lumbar diskectomy" OR "lumbar discectomy" OR "lumbar disk surgery" OR "lumbar disc surgery" OR "lumbar<br>disc" OR "lumbar disk" OR "lumbar disk prolapse" OR "lumbar disc prolapse" OR "prolapsed intervertebral disk"<br>OR "prolapsed intervertebral disc" OR "discogenic compression") AND ("cauda equina syndrome" OR "cauda<br>equina compression" OR "cauda syndrome" OR "cauda equina"[Majr]) |
|--|
| WEB OF SCIENCE (155 results)<br>Same strategy as used in Pubmed, adjusted for this database.   |
| EMBASE (316 results)<br>Same strategy as used in Pubmed, adjusted for this database.   |

Figure 1 Search strategy (performed 22 January 2012)

Two of the authors (NSK, WCHJ) separately screened the articles by title, abstract or by full article, when necessary, to select the studies that met the predefined selection criteria. Selection criteria were stated as followed:

- the article was published in English, Dutch, French, German or Spanish;
- the study included patients diagnosed with cauda equina syndrome due to hernia nuclei pulposi (HNP);
- the study reported the following disease characteristics: HNP diagnosed by means of MRI, CT or X contrast RSG;
- the study reported the following patient characteristics: function of micturition, function of defecation and/or sexual function at base line (e.g. pre-operative) and at follow up (e.g. post operative), with a follow up period of at least two weeks;
- the study included / evaluated a (primary) treatment, excluding reoperations;
- the study was a case study (with a minimum of 10 patients), cohort study or randomized controlled trial. Systematic reviews or meta-analysis were not included;
- the article was published fully in a peer reviewed journal.

Any discrepancy in selection between the two reviewers was resolved in open discussion. Reference screening and citation tracking were performed on the identified articles and as a final check, the meta-analyses found in the first search were studied to make sure no relevant articles were missed.

#### **Quality assessment**

The methodological quality of these studies was assessed by two independent reviewers (HWE, CLAVL), using an adjusted version of the checklist for cohort studies of the Dutch Cochrane Centre.<sup>10</sup> When there was no consensus about the assessment, a third reviewer (NSK) was consulted.

The items reviewed in the assessment were: definition of patient group; selection bias; definition of type of surgery (laminectomy/arcectomy); method for assessing outcome (urodynamic/grading/descriptive) and loss to follow up. A point was given for each of the following items: well-defined patient group (information was given about patients age (mean or range), and about presence/absence of saddle anesthesia, radicular complaints and micturition dysfunction at presentation), absence of selection bias and absence of attrition bias (attrition bias: loss to follow up>20%). Studies were divided into different groups by risk of bias, with the maximum of three points indicating the lowest risk of bias.

### Statistical analysis

Statistical analyses were done in STATA version 11.0.<sup>11</sup> Mean prevalence and range of micturition, defecation and sexual function were calculated. To evaluate the effect of risk of bias and the time of follow up, as well as preoperative proportion of micturition dysfunction, defecation dysfunction and saddle hypo/anesthesia on the amount of dysfunction of micturition, defecation and sexual function at follow up, a meta-regression was performed. Risk of bias was dichotomized in low risk of bias (three points) and medium to high risk of bias (two points or one point). Follow up was dichotomized in twelve months or less and more than twelve months. Backward elimination was used by repeating the test after removing the least significant factors. A Monte-Carlo permutation test was performed to correct for multiple testing and a probability value of <0.05 was considered statistically significant.<sup>12</sup> Sensitivity analyses were performed for influence of shorter follow up and low risk of bias studies.

For studies that also included patients without preoperative dysfunction, we corrected postoperative prevalence of dysfunction for pre-operative prevalence of dysfunction, in order to make comparison between studies fair.

# RESULTS

### Characteristics of included studies and risk of bias

Through our search, 620 articles were identified, of which 527 original articles were left after removing duplicates (Figure 2). Selection procedure and subsequent citation tracking resulted in sixteen reports on fifteen different studies that met all criteria.<sup>5,13-27</sup> One study was reported in two publications.<sup>19,20</sup> Reasons to exclude articles were among others small patient numbers,<sup>28,29</sup> inclusion of HNP patients instead of CES patients exclusively,<sup>30-37</sup> inclusion of hemi CES instead of total CES,<sup>38</sup> no imaging done,<sup>3,4,39-41</sup> no evaluation of a primary treatment,<sup>42-44</sup> follow up of at least two weeks not guaranteed<sup>45-49</sup> and no adequate report on post-operative functions.<sup>50</sup>



Figure 2 Flow chart of literature search

Fifteen studies, published from 1956 to 2011, were included with 464 patients (Table 1). All were retrospective studies with relatively small sample sizes (range 14 to 54) and a rather young patient population (mean 43.5 years). The mean minimal time of follow up was 17.0 months after surgery. Except for three patients who were treated conservatively,<sup>5,17,24</sup> all patients underwent surgery. Four authors did not give exact information about type of surgery.<sup>5,22-24</sup> Outcome was descriptive in eights studies,<sup>13,14,16,17,21,22,26,27</sup> graded in six studies<sup>5,15,18,20,23,25</sup> and both urodynamic and descriptive in one study.<sup>24</sup>

After consultation of the third reviewer, consensus about risk of bias score was achieved in all cases. Six studies were assessed to have a low risk of bias (Table 2). Two studies showed a high risk with both selection bias and either a poor-defined patient group or attrition bias. The remaining seven studies had an intermediate risk of bias.

| Table 1 Characte | ristics     | of included st       | tudies         |   |                              |
|------------------|-------------|----------------------|----------------|---|------------------------------|
| Study            | n<br>(total | Mean age<br>in years | Male<br>gender | Mode of surgery<br>(number of patients)   | Minimal time<br>of follow up |
|                  | 464)        | (range)              | (%)            |   | in months                    |
| Jennett (1956)   | 25          | NR (20-72)           | 60.0           | NR  | 12                           |
| Spännare (1978)  | 30          | NR (20-70)           | 46.7           | Complete laminectomy with discectomy (17), bilateral arcectomy (7), unilateral arcectomy (6)                            | 24                           |
| O'Laoire (1981)  | 29          | 40.6 (23-69)         | 58.6           | Laminectomy with discectomy (29)  | 12                           |
| Hellström (1986) | 17          | 43 (33-63)           | 58.8           | NR (16), no surgery (1)   | 24                           |
| Kostuik (1986)   | 31          | 40 (23-61)           | 54.8           | Wide bilateral laminectomy (30), no surgery (1)   | 24                           |
| Gleave (1990)    | 33          | 40.6 (23-67)         | NR             | Laminectomy (29), fenestration (4)  | 24                           |
| Shapiro (1993)   | 14          | 43 (22-67)           | 64.3           | Laminectomy with discectomy (14)  | 9                            |
| Kennedy (1999)   | 19          | 55 (31-76)           | 63.2           | Bilateral laminectomy (19)  | 22                           |
| Buchner (2000)   | 22          | 42 (22-67)           | 59.1           | Bilateral hemilaminectomy (12), bilateral laminectomy (9)   | 12                           |
| Shapiro (2000)   | 44          | 44 (22-67)           | 61.4           | Laminectomy with discectomy (44) of which 1 unilateral microdiscectomy approach   | 12                           |
| Hussain (2003)   | 20          | 45 (33-67)           | 50.0           | Bilateral laminectomy with discectomy (20)  | 10                           |
| McCarthy (2007)  | 54          | 41 (24-67)           | 54.8           | NR  | 25                           |
| Qureshi (2007)   | 33          | 43 (30-79)           | 57.6           | Interlaminar discectomy (15) of which 6 microdiscectomy approach, laminectomy with discectomy (12), hemilaminectomy (6) | m                            |
| Crocker (2008)   | 43          | NR                   | NR             | NR  | 24                           |
| Dhatt (2011)     | 50          | 48 (25-85)           | 66.0           | Wide laminectomy with discectomy (50)   | 12                           |
| Mean (corrected) |             | 43.5                 | 55.7           |   | 17.0                         |
|                  |             |                      |                |   |                              |

NR = not reported

| Study            | Score on risk of<br>bias scale | Well-defined patient<br>group | Absence of selection bias | Absence of attrition bias |
|------------------|--------------------------------|-------------------------------|---------------------------|---------------------------|
| Jennett (1956)   | ***                            | *                             | *                         | *                         |
| Spännare (1978)  | ***                            | *                             | *                         | *                         |
| O'Laoire (1981)  | ***                            | *                             | *                         | *                         |
| Hellström (1986) | **                             | *                             | -                         | *                         |
| Kostuik (1986)   | **                             | *                             | -                         | *                         |
| Gleave (1990)    | ***                            | *                             | *                         | *                         |
| Shapiro (1993)   | **                             | -                             | *                         | *                         |
| Kennedy (1999)   | **                             | *                             | -                         | *                         |
| Buchner (2000)   | ***                            | *                             | *                         | *                         |
| Shapiro (2000)   | *                              | *                             | -                         | #                         |
| Hussain (2003)   | **                             | *                             | *                         | #                         |
| McCarthy (2007)  | **                             | *                             | *                         | -                         |
| Qureshi (2007)   | **                             | *                             | *                         | -                         |
| Crocker (2008)   | *                              | -                             | -                         | *                         |
| Dhatt (2011)     | ***                            | *                             | *                         | *                         |

#### Table 2 Risk of bias assessment

# no information was provided about exact loss to follow up NB asterisks represent the number of points gained

# **Clinical presentation of CES**

Included studies varied in definition of CES and thus in patient characteristics at presentation (Table 3). Micturition dysfunction was regarded as an important element of CES by most authors and all articles assessed micturition at presentation. Exact definition of micturition dysfunction varied across studies. Mean prevalence of micturition dysfunction at presentation was 88.9%. Eight authors reviewed function of defecation at presentation, resulting in a prevalence of dysfunction in 47.1%. Most authors agreed that saddle anesthesia - or to a lesser extent, saddle hypoesthesia – is one of the hallmarks of the classic presentation of CES. In included studies, saddle anesthesia/hypoesthesia was found in 80.8% of patients at presentation. Pre-operative SD was only assessed in a total of three patients, in three studies.<sup>5,16,22</sup> Sciatica, whether unilateral or bilateral, was present in 95.5% of patients.

| Table 3 Charact   | eristics of patier                              | nts at presentati                              | on  |                               |  |
|-------------------|---|--|---|-------------------------------|--|
| Study             | Micturition<br>dysfunction<br>in % ( <i>n</i> ) | Defecation<br>dysfunction<br>in % ( <i>n</i> ) | Saddle hypo/<br>anesthesia<br>in % ( <i>n</i> ) | Sciatica<br>in % ( <i>n</i> ) | Definitions for micturition dysfunction at presentation  |
| Jennett (1956)    | 92.0 (23/25)                                    | 92.0 (23/25)                                   | 96.0 (24/25)                                    | 100.0 (25/25)                 | "some degree of paralysis of the anal and vesical sphincters"  |
| Spännare (1978)   | 100.0 (30/30)                                   | NR   | 70.0 (21/30)                                    | 100.0 (30/30)                 | "some disorders of micturition"  |
| O'Laoire (1981)   | 100.0 (29/29)                                   | NR   | 51.7 (15/29)                                    | 82.8 (24/29)                  | "impaired sphincter function", i.e. "catheterisation required to treat urinary retention or<br>micturition by bladder compression, either by breath-holding or by manually compressing<br>the abdomen" |
| Hellström (1986)  | 70.6 (12/17)                                    | NR   | 94.1 (16/17)                                    | 100.0 (17/17)                 | "retention of urine", urodynamics  |
| Kostuik (1986)    | 100.0 (31/31)                                   | 54.8 (17/31)                                   | 54.8 (17/31)                                    | 100.0 (31/31)                 | "urinary retention"  |
| Gleave (1990)     | 100.0 (33/33)                                   | NR   | 72.7 (24/33)                                    | 90.1 (30/33)                  | "urinary retention with overflow incontinence", i.e. "inability to void, or bladder enlargement with dribbling incontinence"   |
| Shapiro (1993)    | 92.9 (13/14)                                    | NR   | NR  | 100.0 (14/14)                 | "urine incontinence"   |
| Kennedy (1999)    | 100.0 (19/19)                                   | 78.9 (15/19)                                   | 100.0 (19/19)                                   | 94.7 (18/19)                  | "symptoms of urinary dysfunction"  |
| Buchner (2000)    | 100.0 (22/22)                                   | 36.4 (8/22)                                    | 68.2 (15/22)                                    | 100.0 (22/22)                 | "urinary retention with overflow incontinence requiring catheterization"   |
| Shapiro (2000)    | 100.0 (44/44)                                   | NR   | 100.0 (44/44)                                   | 100.0 (44/44)                 | "urologic problems of retention, incontinence or both"   |
| Hussain (2003)    | 100.0 (20/20)                                   | 70.0 (14/20)                                   | 100.0 (20/20)                                   | 100 (20/20)                   | "lack of bladder control"  |
| McCarthy (2007)   | 59.5 (25/42)                                    | 50.0 (21/42)                                   | 76.2 (32/42)                                    | 90.4 (38/42)                  | "urinary retention"  |
| Qureshi (2007)    | 90.9 (30/33)                                    | 30.3 (10/33)                                   | 81.8 (27/33)                                    | 84.8 (28/33)                  | "urological dysfunction"   |
| Crocker (2008)    | 48.8 (21/43)                                    | NR   | 81.4 (35/43)                                    | 100.0 (43/43)                 | "sphincter distension"   |
| Dhatt (2011)      | 100.0 (50/50)                                   | 12.0 (6/50)                                    | 90.0 (45/50)                                    | NR                            | "minor or major bladder dysfunction"   |
| Mean              | 88.9 (402/452)                                  | 47.1 (114/242)                                 | 80.8 (354/438)                                  | 95.5 (384/402)                |  |
| NR = not reportea |   |  |   |                               |  |

#### Outcome

#### Micturition

Outcome of micturition was evaluated in a total of 409 patients (Table 4). At an average mean minimal follow up time of 17.0 months after surgery, mean prevalence of micturition dysfunction is 42.5% (range 13.3-90.0). The corrected mean prevalence of micturition dysfunction is 45.1% (range 13.3-90.0).

Micturition dysfunction is often defined as incontinence<sup>5,13,14,16,18,19,25</sup> or by the presence of urologic complaints<sup>21,22,24</sup> or disturbances.<sup>15,23,27</sup> Also mentioned are the absence of a 'normal voiding pattern'<sup>17</sup> and 'any residual deficit that was regarded as a physical or psychological impairment'.<sup>26</sup> In one study, in addition to clinical assessment, extensive urodynamics tests were performed to measure micturition outcome, but the author concluded that the complaints patients gave were not always consistent with these functional outcomes.<sup>24</sup>

Variation of prevalence across studies is large. The highest rates of dysfunction were found by Hussain, Dhatt (both 90%) and Jennett (76.9%); these three studies all showed a low to intermediate risk of bias. Lowest prevalences were found by Kostuik and Gleave (13.3% and 21.2%, respectively) which also demonstrated a low to intermediate risk of bias at 24 months post-operative.

#### Defecation

Outcome of defecation function was evaluated in a total of 238 patients.<sup>5,14-16,18,22,24,26</sup> At an average mean minimal follow up time of 17.0 months after surgery, mean prevalence of defecation dysfunction is 49.6%. Prevalences range from 10.5% to 90.0%.<sup>15,26</sup> Interestingly, the studies that evaluated post-operative function of defecation are not the same studies that evaluated this function pre-operatively.

Different definitions of defecation dysfunction are used: 'a patulous anal sphincter leading usually to faecal incontinence' or 'constipation with defective anal sensation',<sup>5</sup> complaints of 'bowel disturbance',<sup>15,22</sup> 'abnormal sphincter tone',<sup>24</sup> being 'grossly incontinent of stool',<sup>14</sup> no control of flatus and being 'occasional incontinence of faeces',<sup>18</sup> 'poor faecal continence and no control of flatulence'<sup>26</sup> and a state different from 'never or rarely leaked from bowels'.<sup>16</sup>

#### Sexual function

Outcome of sexual function was reviewed in a total of 201 patients. Only two studies reviewed sexual function in every patient seen at follow up.<sup>15,17</sup> Seven studies recorded sexual function in a selection of included patients, more often in men than in women.<sup>5,13,14,18,22,24,26</sup> At an average mean minimal follow up time of 17.0 months after surgery, mean prevalence of SD is 44.3% (range 10.0-76.6). Many different varieties of SD are mentioned, even within the same study: impotence,<sup>5,13,14,17,22,24</sup> decreased potency,<sup>14,22,24</sup> more difficult to obtain orgasm,<sup>14</sup> less intense orgasm,<sup>14</sup> anorgasmy,<sup>14,22</sup> decreased<sup>17,22</sup> or absent<sup>14</sup> penile/vaginal sensation, incontinence during intercourse,<sup>14,17</sup> dyspareunia,<sup>22</sup> absent bulbocavernosus reflex,<sup>26</sup> and the very general terms 'SD'<sup>15</sup> and 'abnormal intercourse'.<sup>18</sup>

| Study             | Micturition<br>dysfunction in<br>% (n) | Corrected #<br>micturition<br>dysfunction in<br>% (n) | Defecation<br>dysfunction in<br>% (n) | Corrected #<br>defecation<br>dysfunction in<br>% ( <i>n</i> ) | Sexual<br>dysfunction in<br>% (n) |
|-------------------|--|---|---------------------------------------|---|-----------------------------------|
| Jennett (1956)    | 76.9 (10/13)                           | idem  | 84.6 (11/13)                          | idem  | 25.0 (1/4)                        |
| Spännare (1978)   | 33.3 (10/30)                           | idem  | NR                                    | NR  | NR                                |
| O'Laoire (1981)   | 37.9 (11/29)                           | idem  | 37.9 (11/29)                          | idem  | 35.3 (6/17)                       |
| Hellström (1986)  | 41.2 (7/17)                            | 58.3 (7/12)   | 43.8 (7/16)                           | idem  | 20.0 (2/10)                       |
| Kostuik (1986)    | 13.3 (4/30)                            | idem  | NR                                    | NR  | 26.6 (8/30)                       |
| Gleave (1990)     | 21.2 (7/33)                            | idem  | NR                                    | NR  | NR                                |
| Shapiro (1993)    | 28.6 (4/14)                            | 30.8 (4/13)   | NR                                    | NR  | 75.0 (6/8)                        |
| Kennedy (1999)    | 26.3 (5/19)                            | idem  | 10.5 (2/19)                           | 13.3 (2/15)   | 10.0 (1/10)                       |
| Buchner (2000)    | 22.7 (5/22)                            | idem  | NR                                    | NR  | NR                                |
| Shapiro (2000)    | 36.4 (16/44)                           | idem  | 20.5 (9/44)                           | idem  | 76.6 (23/30)                      |
| Hussain (2003)    | 90.0 (18/20)                           | idem  | NR                                    | NR  | NR                                |
| McCarthy (2007)   | 33.3 (14/42)                           | 56.0 (14/25)  | 59.5 (25/42)                          | 119.0 (25/21)   | 57.1 (24/42)                      |
| Qureshi (2007)    | 44.0 (11/25)                           | idem  | 32.0 (8/25)                           | 80.0 (8/10)   | NR                                |
| Crocker (2008)    | 33.3 (7/21)                            | idem  | NR                                    | NR  | NR                                |
| Dhatt (2011)      | 90.0 (45/50)                           | idem  | 90.0 (45/50)                          | 750.0 (45/6)  | 36.0 (18/50)                      |
| Mean in % (range) | 42.5 (13.3-90.0)                       | 45.1 (13.3-90.0)                                      | 49.6 (10.5-90.0)                      | 76.6 (13.3-750.0)   | 44.3 (10.0-76.6)                  |

Table 4 Outcome of micturition, defecation and sexual function at follow up

#corrected for number of patients with dysfunction at presentation (see Table 3) NR = not reported

#### Sensitivity analysis

We looked more closely at the relationship between follow up time and dysfunction (Table 5). Eight studies had a minimal follow up of twelve months or less (average 9.9 months, range 3-12) and seven studies of more than twelve months (average 23.9 months, range 22-25). In the group with follow up time 12 months or less, prevalence of micturition dysfunction was higher than in the group with follow up time more than 12 months (55.3% versus 28.1%). The difference was statistically significant (p=0.043). Regarding post-operative defecation dysfunction, we found the group with follow up time 12 months or less again showing a higher prevalence of dysfunction than the group with follow up time more than twelve months (52.2% versus 44.2%), this difference was not statistically significant. For sexual function, the difference was statistically

significant (p=0.007), with the group of follow up time with 12 months or less showing a higher prevalence of dysfunction than the group with follow up time with more than 12 months (47.5% versus 38.0%).

| Table B Enect of follow up time on postoperative | eaysianeaon  |  |                              |
|--|--|--|------------------------------|
|  | Studies with<br>minimal follow up<br>time ≤12 months | Studies with<br>minimal follow up<br>time >12 months | Adjusted#<br><i>p</i> -value |
| Postoperative micturition dysfunction in % (n)   | 55.3 (120/217)                                       | 28.1 (54/192)  | 0.043                        |
| Postoperative defecation dysfunction in % (n)    | 52.2 (84/161)  | 44.2 (34/77)   | not significant              |
| Postoperative sexual dysfunction (SD) in % (n)   | 47.5 (48/101)  | 38.0 (35/92)   | 0.007                        |

Table 5 Effect of follow up time on postoperative dysfunction

*# adjusted for multiple testing* 

Reviewing the effect of bias on outcome showed that low risk of bias studies reported higher dysfunction of micturition and defecation at follow up (49.7% and 72.8%, respectively) than the intermediate and high risk of bias studies (37.1% and 34.9%, respectively) (Table 6). The difference for defecation was statistically significant (p=0.017). Regarding sexual function, less dysfunction was seen in the studies with low risk of bias than in the medium-high risk of bias studies (35.2% versus 49.2%), with the difference being statistically significant (p=0.031).

#### Table 6 Effect of risk of bias on postoperative dysfunction

|  | Low risk of bias<br>studies (***) | Medium to high risk of bias studies (*/**) | Adjusted#<br><i>p</i> -value |
|--|-----------------------------------|--|------------------------------|
| Postoperative micturition dysfunction in % (n)       | 49.7 (88/177)                     | 37.1 (86/232)                              | not significant              |
| Postoperative defecation dysfunction in $\%$ ( $n$ ) | 72.8 (67/92)                      | 34.9 (51/146)                              | 0.017                        |
| Postoperative sexual dysfunction (SD) in % (n)       | 35.2 (25/71)                      | 49.2 (64/130)                              | 0.031                        |

# adjusted for multiple testing

# Relation of mode of presentation and reported post-operative dysfunction

Since we would like to know if there is any correlation between pre-operative dysfunction and post-operative dysfunction, e.g., whether patients presented with defecation dysfunction show a higher prevalence of post-operative sexual dysfunction, we evaluated the effect of micturition dysfunction, defecation dysfunction and saddle hypo/anesthesia at presentation on postoperative micturition, defecation and sexual function. However, none of these symptoms were found to be statistically prognostic factors for the outcome of micturition, defecation and sexual function.

#### DISCUSSION

Fifteen studies reviewing outcome of micturition, defecation and sexual function in CES secondary to radiologically confirmed HNP were found, with a minimal mean follow up time of 17.0 months. All reviewed micturition dysfunction (mean prevalence 42.5%), eight studies reviewed defecation dysfunction (49.6%) and nine studies SD (44.3%). Percentages of dysfunction varied largely across studies.

In studies with a shorter follow up time, more dysfunction was reported than in studies with a longer follow up, with a statistically significant difference for micturition and sexual function (p=0.043 and p=0.007, respectively). None of the presenting symptoms analyzed were found to be statistically significant prognostic factors for postoperative micturition, defecation or sexual function.

Lower risk of bias studies reported more dysfunction of micturition and defecation (but less SD; p=0.031), with the difference in defecation dysfunction being statistically significant (p=0.017). We came across several other meta-analyses on various subjects in literature supporting our finding that studies with more risk of bias showed a greater treatment effect.<sup>51-53</sup>

#### Differences in definition and in reviewing outcome

Reviewing literature on outcome for CES involves combining data from various studies. Differences in definitions of CES pose difficulties in comparing outcome between studies. Different definitions of dysfunction across studies (e.g. Table 3) may cause bias when analyzing results together. The obvious solution to this problem is simply to create a clear and workable term for dysfunction; however, this may be more difficult than it sounds. On the one hand we have the physician, looking at dysfunction as organ failure, based on urodynamics and other physical tests, and on the other hand, there is the patient, experiencing dysfunction by complaints and problems in daily life. These two perspectives do not necessarily agree: e.g. in his study McCarthy measured reduced rectal tone in 21 patients, but found complaints of defecation in 25 patients. When a patient is suffering from defecation complaints due to CES, there is a problem of dysfunction, even without measurable dysfunction of normal rectal tone. In addition, the only study that evaluated micturition objectively by means of urodynamics, concluded that the complaints of patients were not always consistent with those tests.<sup>24</sup> Therefore we would like to mark complaints as dysfunction even in the absence of aberrant test results, since it is quite common to not find any dysfunction by objective tests in these cases. The use of standardized questionnaires to ask for complaints, as was done in the study of Kennedy,<sup>26</sup> therefore seems an elegant way to address dysfunction.

#### Sexual function

At an average mean minimal follow up time of 17.0 months after surgery, almost half of patients experience SD. Sexual function may not only be directly, but also indirectly affected by CES which is illustrated by the statement of some patients that incontinence of bladder or bowel often causes distress and a great sense of shame during sexual activity.<sup>14,17</sup>

The assessment of sexual function in included studies seems quite arbitrary and is done more often in men than in women. Perhaps inabilities to obtain or maintain erection are perceived as more basic problems than reduced sensation for women, and the participating clinicians, more often men, may find it easier to discuss sexual function with the same sex. In any case, SD seems a topic difficult to discuss for both doctor and patient. Research on this subject, e.g. in the form of questionnaires for clinicians, can provide information about the place sexual problems have in current therapy, and more specifically, ideas to make the treatment of SD more common practice.

Overall, literature evaluating sexual function after CES is scarce.<sup>54</sup> More is written about the effect on sexual function after HNP<sup>31,55,56</sup> and spinal cord injury,<sup>57-59</sup> some with suggestions for treatment.<sup>60</sup>

# **Overview of literature**

This is the first systematic review done on the outcome of micturition, defecation and sexual function in CES. Reviews have been written about these functions separately, but none of these systematically reviewed outcomes combining these three functions. As stated before, this is partly due to traditional focus of literature on timing of decompression. Two large meta-analyses found statistically significant differences in recovery of, among others, micturition and defecation in favour of early decompression. <sup>6,61</sup> Smaller studies reported similar findings.<sup>13,40,41,50</sup> In one study, the differences were statistically significant.<sup>14</sup> The most recent study that was included in this review evaluated outcome after delayed decompression and stated that the high prevalences of post-operative dysfunction that were found, could possibly have been prevented by early decompression.<sup>15</sup> One of the included studies,<sup>16</sup> together with a study from 2009,<sup>49</sup> did not find differences between outcomes. Other authors suggest emergency surgery is more important for some cases of CES than for others.<sup>62-65</sup> However, no doubt arises that CES is an absolute indication for emergency decompression and surgery should be undertaken as soon as possible to obtain better recovery of functions.<sup>34,41</sup>

#### **CLINICAL INTERPRETATION AND CONCLUSION**

This review offers an insight into the extent of micturition dysfunction, defecation dysfunction and sexual dysfunction in CES after decompression. Since the discussion about timing of decompression has already been researched extensively in literature, instead we looked at the actual outcomes. Our findings show that dysfunction of micturition, defecation and/or sexual function is extremely common with about half of the patients affected in at least one of these domains at an average mean minimal follow up time of 17 months after surgery. So, even though CES patients get decompression as soon as possible with the aim to restore function, a lot of patients still suffer from dysfunction long after surgery, something for which ideally therapy should be provided. Since micturition, defecation and sexual function are closely related and may affect each other, we believe that already existing individual therapies are best to be combined. A condition as invalidating as CES requires good patient information and the outcomes presented in this review may help in providing those data. Bias in included studies, lack of universal definitions and incomplete follow up qualify these data as the best we momentarily have, but still subject to improvement. Since sexual dysfunction (SD) seems to be severely underreported, we recommend further research to explore the extent of this problem, as well as the use of questionnaires next to urodynamic tests in future clinical (prospective) studies to accomplish a more patient-based approach.

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