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

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# CHAPTER 5

## **Cauda equina syndrome: presentation, outcome and predictors with focus on micturition, defecation and sexual dysfunction**

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## ABSTRACT

*Background.* Even though micturition, defecation and sexual function are substantially affected in cauda equina syndrome (CES), data on outcome are scarce.

*Methods.* Medical files of patients operated on lumbar herniated disc were screened for CES and retrospectively analyzed for baseline characteristics, outcome of micturition, defecation and sexual function and possible predictors.

*Results.* Seventy-five CES patients (52% men) were included with a mean age of 44 years. L5-S1 was the most common affected level. Duration of CES complaints at presentation was on average 84 hours (median 48 hours). Prevalence of symptoms at presentation: sciatica (97%), altered sensation of the saddle area (93%), micturition dysfunction (92%), defecation dysfunction (74%). Only 26 patients were asked about sexual dysfunction of whom 25 patients experienced dysfunction. Female gender was associated with more defecation dysfunction at presentation than male gender (OR 4.11;  $p=0.039$ ). All patients underwent decompressive surgery. Two postoperative follow up (FU) moments took place after a mean of 75 hours and 63 days. Outcomes at second FU moment: micturition dysfunction 48%, defecation dysfunction 42%, sexual dysfunction 53%, sciatica 48% and altered sensation of the saddle area 57%. A shorter time to decompression was associated with more sciatica at FU 1 ( $p=0.042$ ) which effect had disappeared at FU 2.

*Conclusion.* This study is unique in (1) displaying the presenting features in a large cohort of CES patients, (2) demonstrating that recovery after decompression is slow and far from complete in the majority of patients with regard to micturition, defecation and sexual function and (3) evaluating predictors for outcome.

## INTRODUCTION

Cauda equina syndrome (CES) is a neurological condition caused by compression of the cauda equina, most commonly described as a combination of sensory loss of the saddle area, motor deficit and/or loss of reflexes of the lower limbs, micturition dysfunction, defecation complaints and/or sexual dysfunction.<sup>1,2</sup> The first article about CES appeared in 1934, in which a combination of neurological and urological complaints in patients with a ruptured intervertebral disc was described.<sup>3</sup> A herniated disc is still the most common cause of cauda equina compression; in literature, 45% of cases of CES are attributed to a lumbar herniated disc.<sup>1</sup>

In the last decades, especially the topic of timing of decompression and its relation to outcome has gained much attention in literature, with several small studies showing better – albeit not always significant – outcomes after early decompression.<sup>4-9</sup> Other studies could not demonstrate a better outcome after early decompression.<sup>10,11</sup> The value of urgent decompression was most convincingly showcased in the meta-analysis of Ahn et al., showing a better prognosis of sensory, motor, urinary and rectal function in patients being decompressed within 48 hours of presentation, compared to the group being decompressed after 48 hours.<sup>12</sup> These results were confirmed by others.<sup>13,14</sup>

In literature, there is little focus on the prognosis of micturition, defecation and sexual function.<sup>2</sup> This is remarkable considering the definition of CES. Recently, clinicians in spinal care were found to barely discuss sexual health and/or defecation at presentation and at follow up, suggested to be due to e.g. lack of knowledge or time.<sup>2,15,16</sup> Clinicians who do want to inform their patients about the prognosis of these functions, are confronted with scarce data. This study was performed in order to (1) evaluate outcome of micturition, defecation and sexual function in CES after decompression and to (2) find possible predictors of outcome. In addition, presenting features of CES were analyzed.

## MATERIAL AND METHODS

The medical records of patients operated in the Leiden University Medical Centre (LUMC; university hospital and referral centre for high complex spinal surgery) between January 1995 and September 2010, with the surgery code 'lumbar discectomy' or 'recurrent lumbar discectomy' were screened by two independent researchers (NSK, JAP) to identify patients with CES. Criteria to diagnose a patient with CES were, according to consensus of literature, one or more of the following: 1) dysfunction of micturition, defecation and/or sexual function (not being attributable to use of opiates or previous disease), 2) altered sensation of the saddle area, with possible neurologic deficit in the lower limb (motor or sensory loss or reflex changes).<sup>1,2</sup> Patients filed with a diagnosis of CES but not

meeting those criteria were excluded. In case of doubt about the diagnosis of CES, a third assessor (CLAVL) was consulted. In order to check inter observer reliability between the two reviewers, 10% of cases were independently screened by both of them.

The following data were extracted from the medical file:

- Baseline characteristics (at presentation): gender; age; level of herniated disc as stated in the file; relevant medical history; referring centre (if applicable); use of opiates and/or laxatives; duration of complaints of herniated disc (defined by the presence of sciatica); duration of CES complaints; information about micturition, defecation, sexual function, altered sensation of the saddle area and/or sciatica; information about anal sphincter reflex and anal sphincter tension.
- Surgery: time between presentation to first doctor and decompression (time to decompression); type of decompressive surgery.
- Follow up: information about micturition, defecation, sexual function, altered sensation of the saddle area and/or sciatica at three follow up moments: (1) at hospital discharge (FU 1; in case notes were taken several times during the first days post-operative, the last notes before discharge were used); (2) at the first outpatient visit, regularly planned 6 weeks postoperatively (FU 2); (3) at the second outpatient visit, which was not regularly planned (FU 3).

Data were collected in Excel and imported in SPSS.

### **Statistical analyses**

Data were analyzed using SPSS Statistics 23.0 (SPSS Inc., Chicago, IL, USA). Demographic values and other patient characteristics were analyzed with frequencies. Investigating proportions between independent groups of categorical data was done with Pearson's Chi-squared test; Fisher Exact Test was used to compare groups with cell counts less than expected. For paired groups with categorical data, McNemar's test was used. Predictors for outcome and presentation were analyzed using a binary logistic regression model; in case of quasi-complete separation of the data, the concerning model was not run or the concerning predictor was removed from the model in order to maintain reliable models. Two-sided  $p$ -values  $< 0.05$  were considered statistically significant. In case of multiple testing, the Bonferroni method was used to correct  $p$ -values. Some numerical data were grouped together for analyses, e.g. timing of decompression was stratified into 6 groups:  $\leq 12$  hours, 13-24 hours, 25-36 hours, 37-48 hours, 49-72 hours and  $> 72$  hours.

### **Missing data**

To run the regression models and for displaying outcome at FU 2, multiple imputation with five imputation sets was used for the following variables: duration of CES complaints; duration of complaints of herniated disc; time to decompression; micturition

dysfunction at FU 1, 2 and 3; defecation dysfunction at presentation, FU 1, 2 and 3; altered sensation of the saddle area at FU 1, 2 and 3; sciatica at FU 1, 2 and 3. Multiple imputation was not performed for sexual dysfunction data due to scarcity of these data, to avoid bias. Pooled data (i.e. derived through multiple imputation) are presented as main data. Original data (not derived through multiple imputation) are presented as corrected (corrected for the number of patients for whom documentation is available at the concerning follow up moment) and as raw (not corrected for the number of patients). Due to an anticipated high amount of loss to follow up at FU 3, outcome at FU 2 was defined as main outcome and data at FU 3 were not used for regression analysis. For the patients for whom data at FU 3 are available, this will be mentioned separately.

## RESULTS

In the period January 1995-September 2010, a total of 744 surgeries coded as '(recurrent) lumbar discectomy' were performed at LUMC, for a total of 696 patients: 38 patients had surgery twice, 10 patients had triple surgery. Out of 696 patients, 75 patients (10.8%) were found to have CES. One female patient who underwent a lumbar discectomy twice, met CES criteria twice; however, since she had not recovered from her first CES completely, only the first surgery was included for analysis. Inter observer reliability regarding diagnosing CES was analyzed using Cohen's Kappa. There was substantial agreement between the two reviewers' judgement ( $\kappa=0.635$ ).

### Patient characteristics

For patients characteristics; see Table 1 and Figure 1. Since the LUMC serves as a referral hospital, the majority of included patients presented first at other hospitals (73.7%). Thirty-two patients (42.7%) used opiates at presentation. Use of laxatives was not significantly higher in the group using opiates: 25.0% of opiate positive patients used laxatives versus 19.5% of opiate negative patients ( $p=0.574$ ).

### Presenting features of CES

Information about sciatica, altered sensation of the saddle area and micturition dysfunction was available for all patients at presentation and was present in 97.3%, 93.3% and 92.0%, respectively (Figure 2). Majority of sciatica was unilateral (60.3%). Altered sensation of the saddle area was classified as either hypoesthesia (75.7%), anesthesia (17.1%) or dyesthesia (7.1%).

Micturition dysfunction was classified as having an indwelling catheter (39.1%), needing clean intermittent catheterization (2.9%), documented residual of bladder (5.3%) or subjective complaints, e.g. reduced feeling of passing urine or mild incontinence (52.2%).

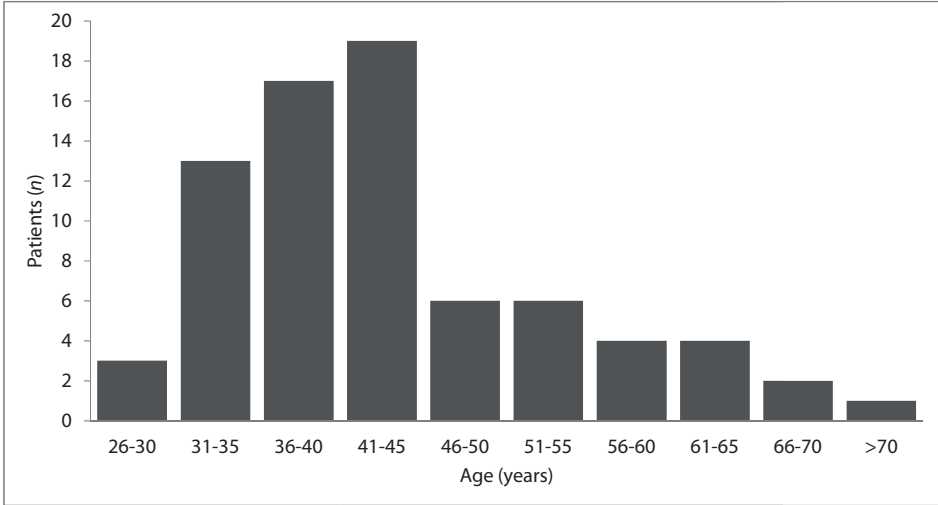
<b>Table 1</b> Patient characteristics ( <i>n</i> =75)		<i>n</i> (%)
<b>Male</b>		39 (52.0)
<b>Mean age</b>	43.6 years (SD 10.4, range 27-78)	
<b>Level of disc lesion (as documented in file)*</b>		
L1-L2		1 (1.3)
L2-L3		3 (3.8)
L3-L4		6 (7.6)
L4-L5		29 (36.7)
L5-S1		39 (49.3)
L6-S1		1 (1.3)
<b>Origin of referral</b>		
Neurologist LUMC		15 (20.0)
Emergency Room LUMC		2 (2.7)
Other hospital in the area		55 (73.3)
General practitioner		2 (2.7)
Unknown		1 (1.3)
<b>Use of opiates at presentation</b>		
Yes		32 (42.7)
No		41 (54.6)
Unknown		2 (2.7)
<b>Use of laxatives</b>		
Yes		16 (21.4)
No		58 (77.3)
Unknown		1 (1.3)
<b>Median duration of herniated disc complaints at presentation (<i>n</i>=73)</b>	30 days (range: 1 day – 14 years)	
<b>Median duration of CES at presentation (<i>n</i>=68)</b>	48 hours	

\*total disc lesions: 79 (4 patients had double lesions: L4-L5+L5-S1 (*n*=3) and L2-L3+L4-L5)

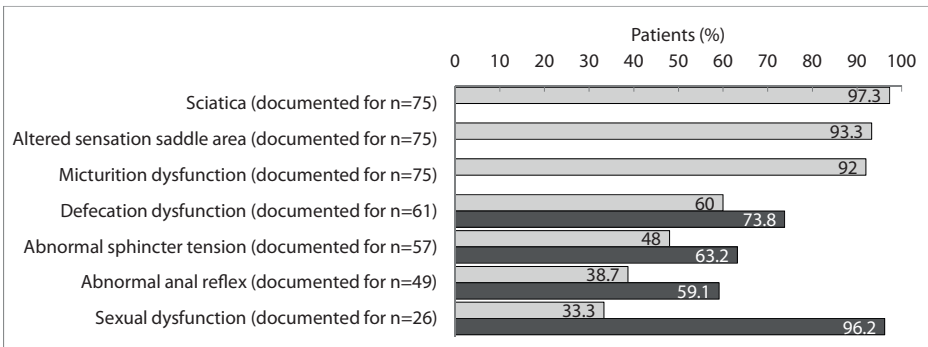
Information about the presence of defecation dysfunction was available in 61 patients, of whom 73.8% had dysfunction, classified as any complaint of defecation which did not exist before, which could be e.g. incontinence or changed sensation of passing stool. A patient with faecal incontinence since diagnosis of M. Sjogren several years before CES, was not classified as having complaints of defecation due to CES.

Information about presence of sexual dysfunction was available for 26 patients (19 men), of whom 25 experienced sexual dysfunction. Documented problems were e.g. altered sensation of genitals, inability to reach orgasm, erectile dysfunction and priapism. For two patients, sexual status (active/non active) before onset of CES was documented; for the others, no notes on sexual activity were found.





**Figure 1** Age distribution of CES patients at time of surgery



**Figure 2** Prevalence of signs and symptoms of CES at presentation. The grey bars indicate the proportion of total patients included in this study ( $n=75$ ); the black bars indicate the proportion of patients for whom documentation was available ( $n$  specified after each row).

Anal sphincter tension and anal sphincter reflex (anal wink) were tested in 76.0% and 65.3% of patients respectively, and were abnormal in the majority (63.2% and 59.1%, respectively). Abnormal anal sphincter tension was significantly associated with altered sensation of the saddle area ( $p=0.007$ ; Table 2), with a sensitivity for altered sensation of the saddle area of 68%. Abnormal anal sphincter reflex was not significantly associated with defecation dysfunction, although a trend was observed ( $p=0.096$ ; Table 3). Micturition dysfunction was not associated with either abnormal anal sphincter tension nor reflex (Table 4). Since data on sexual dysfunction at presentation were scarce, no analyses were done for sexual dysfunction.

**Table 2** Association between altered sensation of the saddle area and sphincter tests. Proportion of patients with abnormal sphincter tests, stratified by sensation of the saddle area (at presentation).

	Altered sensation saddle	Normal sensation saddle	<i>p</i> -value
Abnormal anal sphincter reflex	60.9%	33.3%	0.347
Abnormal anal sphincter tension	68%	0%	0.007

**Table 3** Association between defecation dysfunction and sphincter tests. Proportion of patients with abnormal sphincter tests, stratified by defecation dysfunction (at presentation).

	Defecation dysfunction	No defecation dysfunction	<i>p</i> -value
Abnormal anal sphincter reflex	64.5%	33.3%	0.096
Abnormal anal sphincter tension	63.9%	58.3%	0.731

**Table 4** Association between micturition dysfunction and sphincter tests. Proportion of patients with abnormal sphincter tests, stratified by micturition dysfunction (at presentation).

	Micturition dysfunction	No micturition dysfunction	<i>p</i> -value
Abnormal anal sphincter reflex	60.9%	33.3%	0.347
Abnormal anal sphincter tension	63.5%	60%	0.878

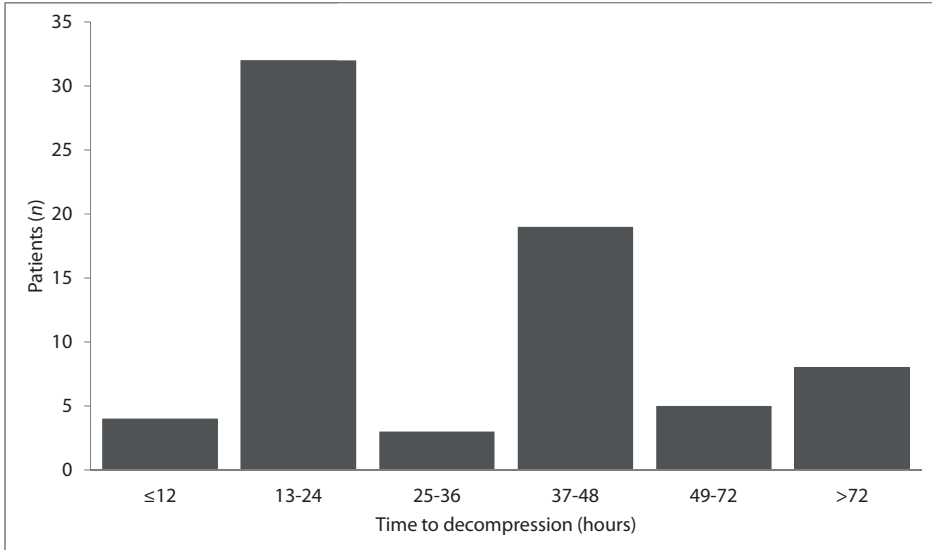
### Association patient characteristics – presenting features

The following factors were evaluated as predictors for presentation: age, gender, duration of complaints of herniated disc, duration of CES complaints, presence of altered sensation of the saddle area (for evaluating defecation and micturition dysfunction at presentation), presence of micturition dysfunction (for evaluating defecation dysfunction and altered sensation of the saddle area at presentation), presence of defecation dysfunction (for evaluating micturition dysfunction and altered sensation of the saddle area at presentation). For sciatica at presentation, the regression model could not be run, nor could sciatica at presentation be added as a predictor to the other models due to quasi-separation of the data. Defecation dysfunction at presentation was significantly associated with female gender (OR 4.11;  $p=0.039$ ). Micturition dysfunction and altered sensation of the saddle at presentation displayed no predictors.

### Surgery

All patients were decompressed by (partial) laminectomy and subsequent discectomy or sequesterectomy. A slight majority of patients ( $n=36$ ) was decompressed within 24 hours after presentation (Figure 3). Eight patients were decompressed more than 72 hours after presentation to the first doctor, with time to decompression of 96 hours ( $n=3$ ), 120 hours ( $n=1$ ), 138 hours ( $n=1$ ), 168 hours ( $n=1$ ), 192 hours ( $n=1$ ) and 216 hours ( $n=1$ ). In 7 of these cases, majority of the delay was caused by the first doctor (family doctor or neurologist) where the patient presented. Hereafter surgery was performed

within 24 hours ( $n=4$ ), 48 hours ( $n=2$ ) and 72 hours ( $n=1$ ) after first presentation to the neurosurgeon. In one case, no discrimination could be made between delay at first and second doctor.



**Figure 3** Time to decompression, counted from the first doctor visit until surgery ( $n=71$ )

### Postoperative outcome

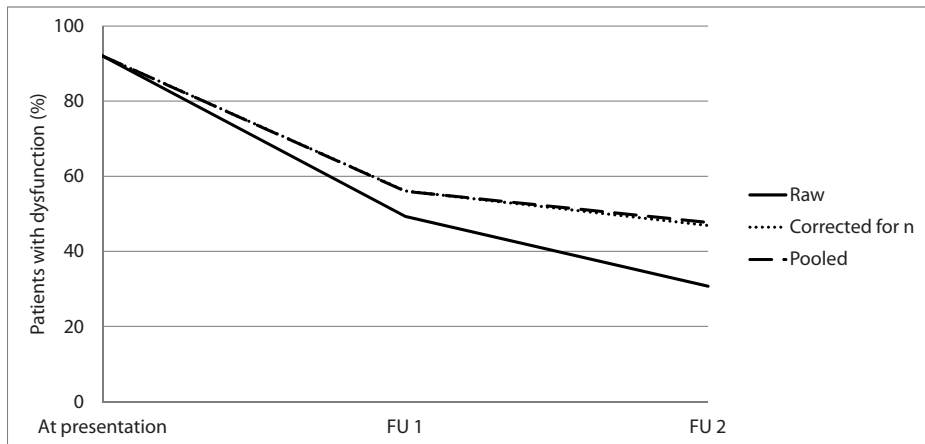
The first follow up moment (FU 1) at which micturition, defecation, sexual function and/or altered sensation of the saddle area was documented was on average 75 hours post-operatively (range 4-336 hours; median 48 hours). The latest time of FU 1 was 14 days post-operatively. Documentation on any of the items micturition, defecation, sexual function and/or altered sensation of the saddle area at the second follow up moment (FU 2), was available for 54 patients (72%), with a mean FU time of 63 days (range 4-300 days, median 60 days).

A third follow up moment (FU 3) at any item was documented for 23 patients (31%), with a mean FU time of 265 days (range 56-730 days, median 225 days). FU 3 is reported in the text as corrected, not pooled, and is not used in any regression analysis.

### Micturition

Documented micturition dysfunction decreased significantly comparing pre-operative moment with FU 2 (92.0% versus 47.7%,  $p<0.001$ ), Figure 4. In one patient with post-operative dysfunction, requiring intermittent catheterization from the 4<sup>th</sup> day after surgery for a short period of time with complaints of urinary dysfunction up to the last follow up moment, (6 months later), no complaints of micturition were documented at

presentation. This was interpreted as misinformation at baseline. Pooled prevalence of micturition dysfunction was 47.7% at FU 2. Reported dysfunction included: indwelling catheter, on-off catheterisation, suprapubic catheter, reduced feeling of passing urine and (mild) incontinence. At FU 3, 19 patients were evaluated of whom 11 displayed dysfunction (57.9%).



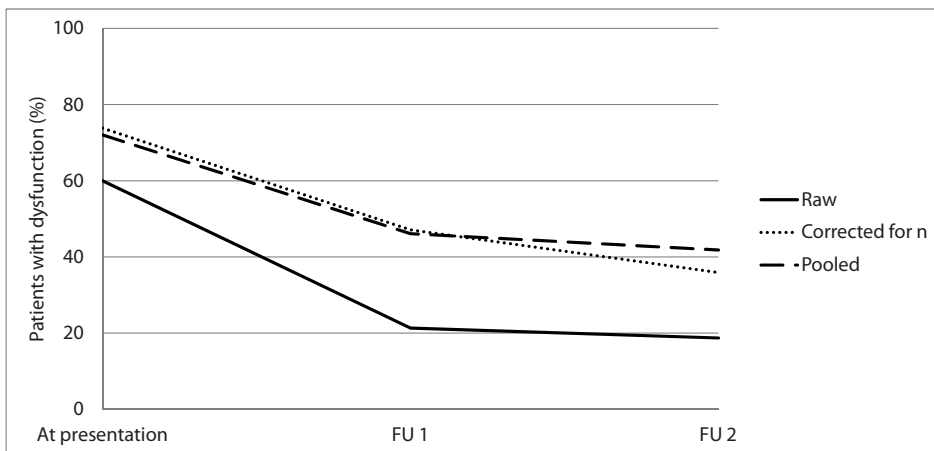
**Figure 4** Outcome of micturition dysfunction. Proportion of patients with micturition dysfunction at presentation (documented for  $n=75$ ), FU 1 (documented for  $n=66$ ) and FU 2 (documented for  $n=49$ ).

### Defecation

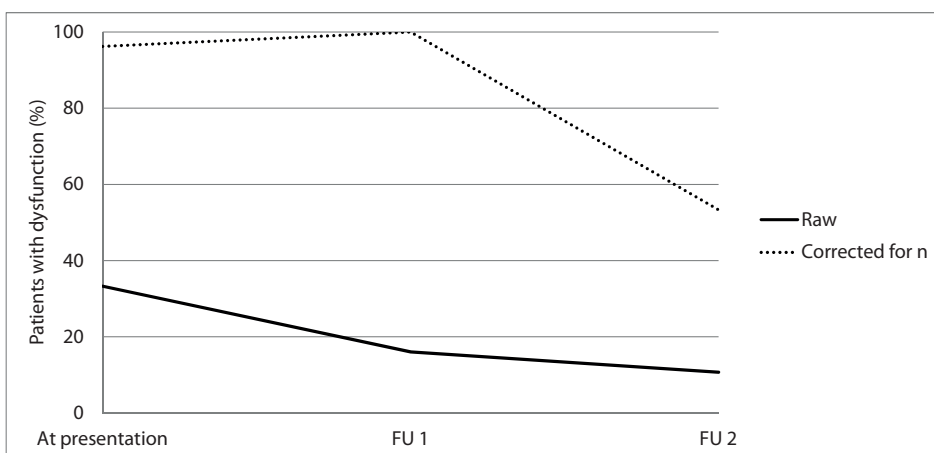
Documented defecation dysfunction had decreased significantly after decompression measured at FU 2 (72.0% versus 41.8%,  $p=0.004$ ), Figure 5. For three patients, defecation dysfunction was documented post-operative but not pre-operative. Pooled prevalence of dysfunction was 41.8% at FU 2. Thirteen patients were evaluated at FU 3, of whom 9 reported dysfunction (69.2%).

### Sexual function

Due to scarce data on sexual function, investigating proportions was not done; therefore, no  $p$ -value for difference between pre- and postoperative dysfunction was derived. Corrected prevalence of sexual dysfunction at FU 2 was 53.3%, Figure 6. Documentation on sexual dysfunction was done for 5 patients at FU 3; four of them displayed dysfunction (80%).



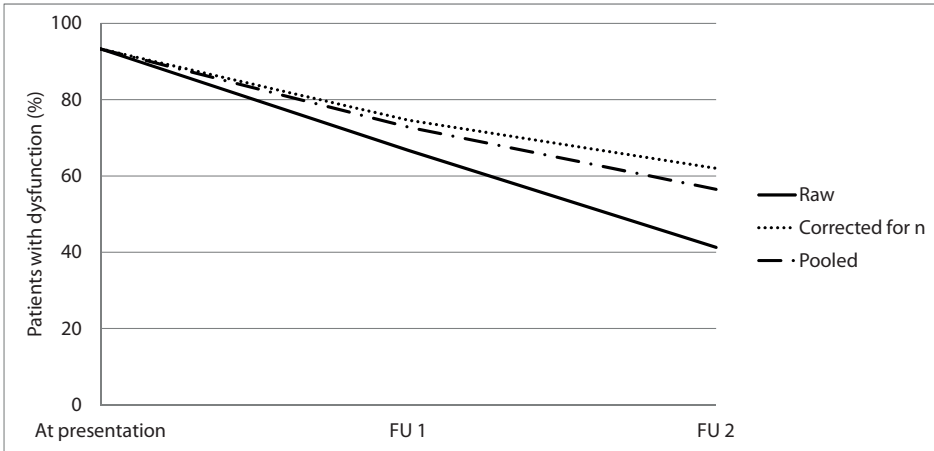
**Figure 5** Outcome of defecation dysfunction. Proportion of patients with defecation dysfunction at presentation (documented for  $n=61$ ), FU 1 (documented for  $n=34$ ) and FU 2 (documented for  $n=39$ ).



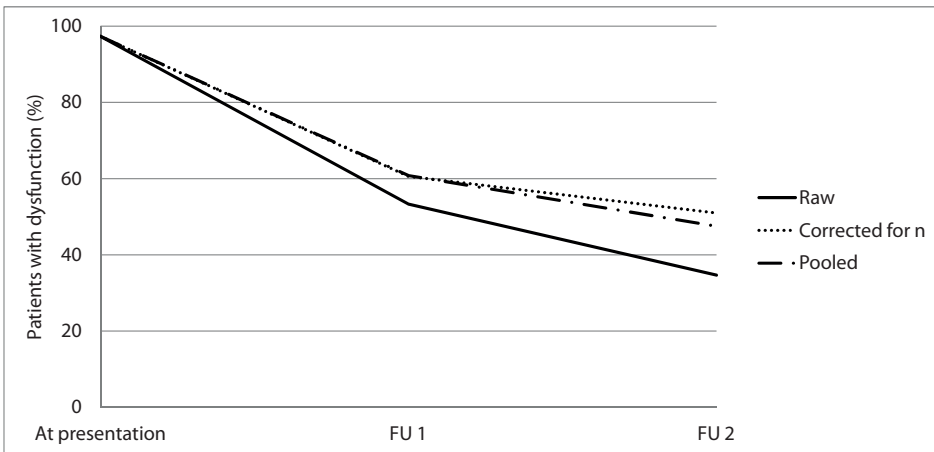
**Figure 6** Outcome of sexual dysfunction. Proportion of patients with sexual dysfunction at presentation (documented for  $n=26$ ), FU 1 (documented for  $n=12$ ) and FU 2 (documented for  $n=15$ ). Since multiple imputation was not used for data of sexual dysfunction, pooled data are not available.

*Altered sensation of the saddle area and sciatica*

Documented altered sensation of the saddle area and sciatica both decreased significantly after decompression measured at FU 2 (93.3% versus 56.5% and 97.3% versus 47.5%, respectively; both  $p<0.001$ ), Figure 7 and 8. At FU 3, 12 out of 18 patient with documentation reported altered sensation of the saddle area (66.7%), and 12 out of 20 reported sciatica (60%).



**Figure 7** Outcome of altered sensation of the saddle area. Proportion of patients with altered sensation of the saddle area at presentation (documented for  $n=75$ ), FU 1 (documented for  $n=67$ ) and FU 2 (documented for  $n=50$ ).



**Figure 8** Outcome of sciatica. Proportion of patients with sciatica at presentation (documented for  $n=73$ ), FU 1 (documented for  $n=66$ ) and FU 2 (documented for  $n=51$ ).

### Predictors for outcome

The following factors were evaluated as predictors for outcome: age, gender, duration of complaints of herniated disc, duration of CES complaints, time to decompression (stratified groups, see Methods section for details of groups), altered sensation of the saddle area/micturition dysfunction/defecation dysfunction/sciatica at presentation.

Due to quasi-separated data, sciatica at presentation had to be removed from all models except the one for micturition dysfunction at FU 2; altered sensation of the saddle

at presentation had to be removed from all models except the one for sciatica at FU 1; micturition dysfunction at presentation had to be removed from the models for sciatica at FU 2 and altered sensation of the saddle area at FU 1 and FU 2.

Timing of decompression was found to be significantly associated with short term outcome (FU 1) of sciatica: less time to decompression was associated with more sciatica at FU 1 ( $p=0.042$ ). After stratifying outcomes for time to decompression using the well known break points from literature of 48 hours<sup>12,14</sup> and 36 hours<sup>13</sup>, we found a statistically significant difference for outcome of sciatica at FU 1 comparing decompression before and after 36 hours. Patients decompressed within 36 hours experienced more sciatica than patients decompressed after 36 hours (79.4% versus 37.9%, adjusted  $p=0.032$ ). There was no difference comparing patients being decompressed before and after 48 hours. There was no association between timing to decompression and outcome of sciatica at FU 2 ( $p=0.475$ ). No other statistically significant predictors for micturition, defecation, altered sensation of the saddle area and sciatica at FU 1 and FU 2 were identified.

## DISCUSSION

This is the largest single study performed about outcome of micturition, defecation and sexual function in CES ( $n=75$ ). The incidence of CES among patients being operated for herniated disc was relatively high in this study: 10.8% compared to 1-3% in literature.<sup>9,17</sup> This high incidence can be explained by the fact that the LUMC serves as a referral hospital for urgent neurosurgical cases. The CES definition that was used to include patients in this study is widely used in literature, and even though a univocal definition for CES does not exist, the authors believe that using this definition guaranteed a fair representation of CES patients. This study displays unique data on the presenting symptoms of a large group of CES patients, proving that, next to the well acknowledged micturition dysfunction, also defecation and sexual dysfunction are common at presentation.

Anal sphincter tension and anal sphincter reflex are often tested in CES patients, even though several studies found no diagnostic value for these tests.<sup>18-21</sup> In the current study, abnormal anal sphincter reflex was not significantly associated with any of the diagnostic criteria for CES used in this study (closest to an association was defecation dysfunction with  $p=0.096$ ). Abnormal anal sphincter tension is significantly associated with altered sensation of the saddle area ( $p=0.007$ ). Specificity of the test is 100% (all patients without altered sensation of the saddle area displayed normal sphincter tension) and sensitivity is 68% (a substantial proportion of patients with anamnestic altered sensation of the saddle area displayed normal sphincter tension). With a positive predictive value of 100% (all patients with abnormal sphincter tension had anamnestic altered

sensation of the saddle area) and a negative predictive value of merely 19%, abnormal anal sphincter tension at physical examination supports the presence of altered sensation of the saddle area, but in no way rules out altered sensation of the saddle area in case it is normal. Considering a specificity of 100%, the question arises whether it is necessary to test sphincter tension in patients without altered sensation of the saddle area since it might cause unnecessary discomfort. Evaluating the anal sphincter reflex data, however, does indicate that sphincter testing might add extra information. Of the five patients with normal sensation of the saddle area at presentation, four were tested for anal sphincter tension (all normal). Two of those four were also tested for the anal sphincter reflex, which in one case was abnormal, demonstrating that with normal sensation of the saddle area and with normal sphincter tension, the anal sphincter reflex can still be abnormal. It could therefore be sensible to do anal sphincter reflex tests even in a patient with normal sensation of the saddle area. The fact that only a small proportion in our study sample demonstrated normal sensation of the saddle area at presentation ( $n=5$ ), limits our data in this aspect and therefore clinical relevance of sphincter tests cannot be secured nor refuted based on those findings.

Prevalence of micturition dysfunction is 47.7% of patients at FU 2. In an older study discussing 13 patients with CES due to herniated disc, the author states that “in all such patients, there was incomplete return of normal micturition”.<sup>22</sup> In a more recent study of McCarthy et al., better recovery of micturition has been described: of 42 evaluated patients with CES due to herniated disc, 36% reported urinary incontinence (mean FU time of 60 months).<sup>23</sup> The higher prevalence of micturition dysfunction in the current study might be due to the shorter FU time, assuming that function of micturition will improve gradually over time.

Regarding defecation dysfunction, this study found a prevalence of 41.8% at FU 2; McCarthy et al. found a higher prevalence at follow up (60%): this could be due to attrition bias, the chance of which becomes greater when follow up period of the study is longer – as in McCarthy’s study. Sexual dysfunction in the current study was 53.3% at FU 2, which is quite similar to the prevalence of 57% reported by McCarthy et al. The true prevalence of sexual dysfunction might be higher than the ones found in studies; since the threshold to discuss sexual health is very high, it is unlikely that either doctor or patient opens the topic, even if there are complaints.

Two patients in this study displayed a specific feature of sexual dysfunction: priapism. One patient reported spontaneous erections at presentation; unfortunately, no documentation on sexual function was done at follow up for this patient. The second patient presented with a numb feeling of the penis which had changed to priapism at follow up after 7 weeks and after 5.5 months. Priapism as a feature of cauda equina compression is extremely uncommon and only two reports in literature describe such a case: one involves a 61-year old man with a herniated disc at L4-L5,<sup>24</sup> the other a 60-year old man with



a degenerative stenosis at L3-L4 and lumbar arachnoiditis.<sup>25</sup> Both patients experienced priapism and a numb respectively a burning sensation at the saddle area when walking, without sphincter disturbances. After decompression of the cauda equina, both patients experienced immediate and complete relieve of their symptoms, suggesting a causal relationship of cauda equina claudicatio and priapism. The parasympathic fibres that are responsible for penile erection arise in S2-S4, and it is thought that their stimulation through (in these two cases: intermittent) compression, had resulted in priapism.<sup>24</sup> To the authors' best knowledge, there are no case reports about priapism in non intermittent cauda equina compression. Even though the course of priapism complaints of the two patients in this study are uncertain, the authors believe that it is not unthinkable that the priapism experienced by the two patients in this study might be attributed to compression of the cauda equina, even though exact mechanisms remain unclear.

This study found that female patients are more likely to present with defecation dysfunction than their male counterparts (OR 4.11;  $p=0.039$ ). Epidemiologic studies demonstrate that female gender is associated with more constipation, as a baseline fact in the general population as well as in the CES population when focusing on outcome.<sup>26,27</sup>

Sciatica were more often present at FU 1 when time to decompression was shorter (OR 1.86;  $p=0.042$ ), which association had disappeared at FU 2. This association nowise undermines the importance of emergency decompression. It rather displays a correlation between duration of compression and other (prognostic) factors for which could not be corrected. Those factors are (1) the speed with which the compression has arisen, stating that patients with slowly developing anatomical lesions have a more favourable prognosis,<sup>28,29</sup> and (2) the type of CES lesion, stating that a "complete" CES (with total obstruction) carries a poorer prognosis than an "incomplete" CES, adding that the incomplete type often occurs more gradually (although not exclusively).<sup>22,30</sup> It seems sensible that a patient with sudden onset of heavy symptoms in general presents earlier. This results in a shorter duration of CES complaints (patient delay) as well as a shorter time to decompression (doctor delay), even though outcome is poor (due to the extent of the lesion). Patients that show reasonable outcome when decompression is delayed by weeks,<sup>31</sup> probably had a favourable anatomical lesion and more gradual onset of complaints. Since it is not (yet) possible to distinguish the group with the favourable conditions from the group with the less favourable conditions at the time of presentation (since e.g. the exact correlation of clinical presentation and degree of canal obstruction on imaging is not yet known) it is necessary to decompress every CES patient as soon as possible, to create the best chances for fair recovery.

The authors strongly believe that this study's finding, e.g. that time to decompression is not associated with outcome of micturition, defecation and altered sensation of the saddle area, and does display a correlation of more sciatica at FU 1 when it is shorter, does not implicate that decompression in CES is to be delayed. Firstly, the number of

patients with delayed decompression in this study is relatively small (eight patients in the group decompressed after 72 hours, five patients in the group decompressed within 48-72 hours). Previously, meta-analyses have displayed better outcomes with decompression taking place within 48 hours<sup>12,14</sup> or within 36 hours.<sup>13</sup> Some studies with smaller patient numbers display a significant better outcome after earlier decompression as well, with significant better outcomes with decompression within 48 hours<sup>9</sup> and even within 10 days.<sup>4</sup> Studies displaying no difference in outcome are a minority and evaluate relative small patient numbers only.<sup>10,11</sup> The finding in this study that a shorter time to decompression is associated with more sciatica at FU 1, should therefore be weighed by the small patient number of this study and the fact that this finding is not present at FU 2 anymore. Also, in this study, clinical motives – unknown to the authors due to the retrospective study design – could have led to the decision for very early decompression in specific patient groups, which might have caused selection bias. The outcome would then be more influenced by factors on which clinical motives are based (and which have led to a an early time to decompression) than by the actual time to decompression.

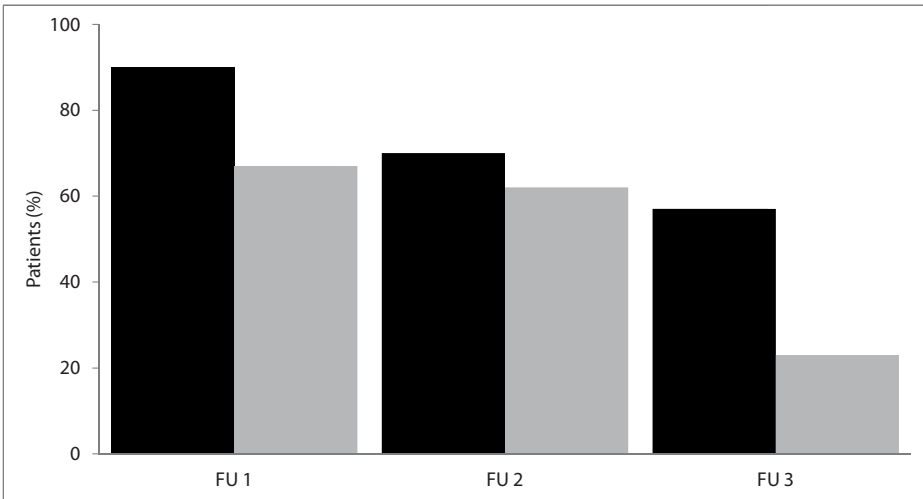
Significant predictors such as duration of CES complaints for more than 48 hours as a risk factor for micturition dysfunction<sup>32</sup> and defecation dysfunction at presentation as a risk factor for sexual dysfunction<sup>23</sup> could not be identified in this study.

Missing data in this study are partly attributed to the inclusion of patients that were referred for surgery to LUMC, and were seen for follow up at their original referring hospital (in which case the researchers did not have access to the follow up data). Of the 19 patients that originated from either LUMC or the general practitioner, 100% displayed data at FU 2. Of the 55 patients that originated from a different hospital and were referred to LUMC for surgery, 34 patients (61.8%) displayed data at FU 2. The amount of dysfunction between the group of LUMC patients and the group originating from a different hospital and referred to LUMC, was not significantly different at FU 2: neither for micturition dysfunction (42.1% versus 48.3%;  $p=0.771$ ) nor for defecation dysfunction (23.5% versus 45.5%;  $p=0.193$ ), sciatica (57.9% versus 46.9%;  $p=0.565$ ) or altered sensation of the saddle area (58.8% versus 63.6%;  $p=0.767$ ). This reason for missing data was therefore not believed to have caused bias.

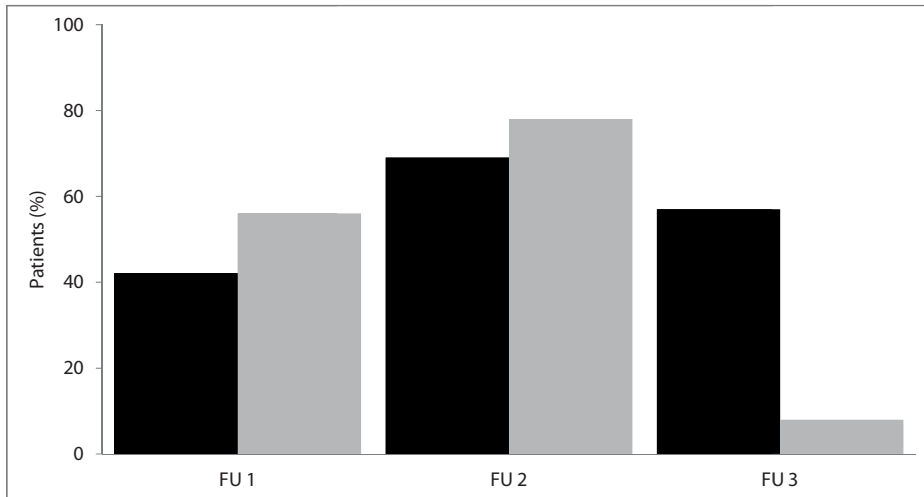
Apart from loss to follow up of patients belonging to a different original hospital, three other reasons for missing data in this retrospective design are: (1) the topic was not discussed and therefore no notes are available, (2) the topic was discussed, but no notes were taken, (3) the patient did not show up at FU moment. Especially for the latter two reasons, data is more likely to be missing when there are no complaints. To explore the extent of this bias – i.e. the hypothesis that data of patients with no complaints are more prone to be missing – patients with and patients without documented complaints at previous check-up were analyzed for the amount of available data (Figures 9-13). As is displayed, patients without documented complaints at their previous check-up, are

more prone to have missing data (apart from the case of defecation dysfunction, which cannot be easily explained). Due to this fact, the authors have chosen to not use FU 3 as main outcome parameter, even more since FU 3 is more likely to be planned for patients with complaints, therefore contributing substantially to attrition bias when it would be used as a main outcome parameter.

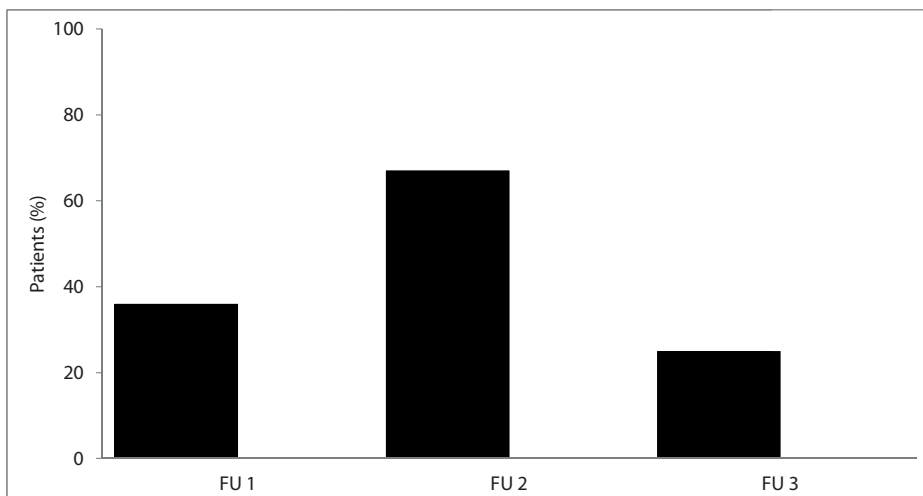
Data on sexual function in this study was particularly scarce, as well at presentation as during follow up, which resulted in the inability of performing several analyses regarding sexual dysfunction. Limited data on sexual function is ubiquitous in CES patients in literature,<sup>2</sup> most likely due to barriers on both the patients side as well as on the doctor's side, which could unfortunately not be minimized in this retrospective study design. The nature of the available data on sexual dysfunction at presentation is striking: 25 out of 26 patients experienced sexual dysfunction. The fact that sexual function is more often documented for male patients than for their female counterparts is something that is believed by the authors to be due to both patient factors as well as doctor factors.<sup>15</sup>



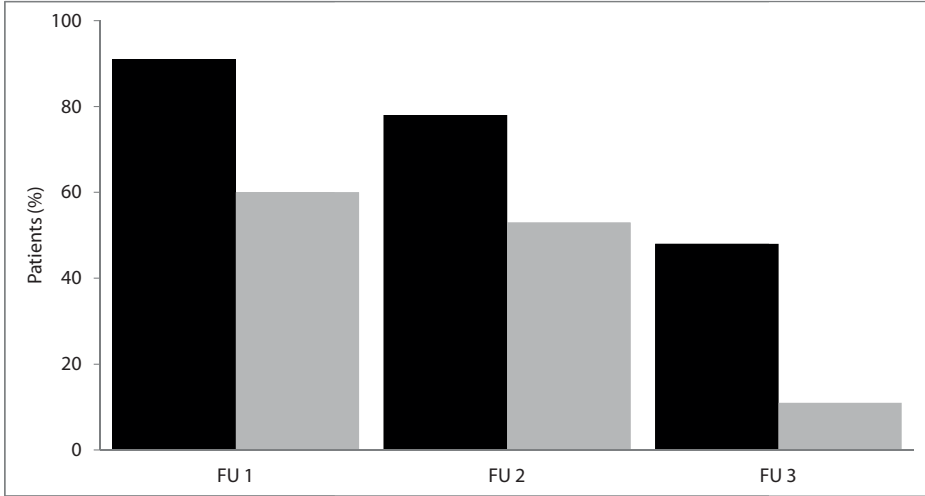
**Figure 9** Availability of data on micturition. The proportion of available data per FU moment is stratified by patients for whom complaints were documented at the previous FU moment (black bar) and by patients for whom it was documented that there were no complaints at the previous FU moment (grey bar).



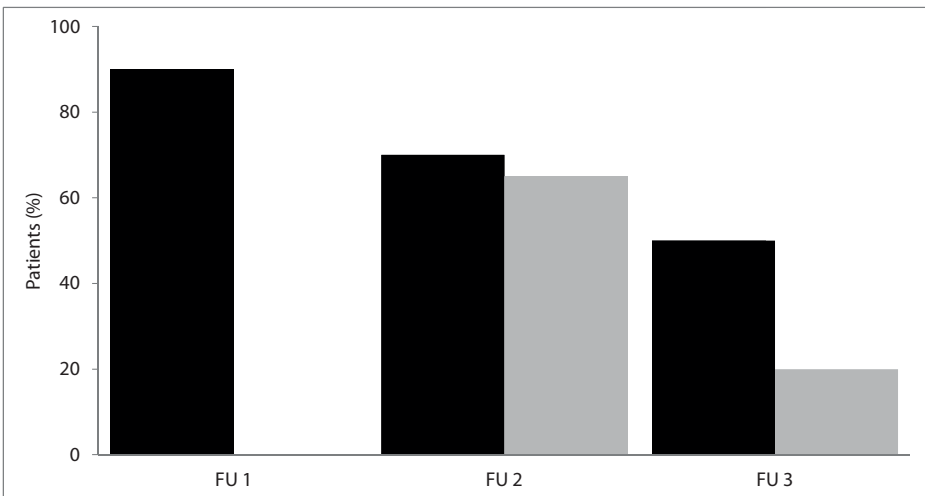
**Figure 10** Availability of data on defecation. The proportion of available data per FU moment is stratified by patients for whom complaints were documented at the previous FU moment (black bar) and by patients for whom it was documented that there were no complaints at the previous FU moment (grey bar).



**Figure 11** Availability of data on sexual function. The proportion of available data per FU moment is shown for patients for whom complaints were documented at the previous FU moment (black bar). For all patients for whom it was documented that there were no complaints at the previous FU moment, there was no data available at the next FU moment, therefore, there are no grey bars.



**Figure 12** Availability of data on sensation of the saddle area. The proportion of available data per FU moment is stratified by patients for whom complaints were documented at the previous FU moment (black bar) and by patients for whom it was documented that there were no complaints at the previous FU moment (grey bar).



**Figure 13** Availability of data on sciatica. The proportion of available data per FU moment is stratified by patients for whom complaints were documented at the previous FU moment (black bar) and by patients for whom it was documented that there were no complaints at the previous FU moment (grey bar). For all patients for whom it was documented that there were no complaints at presentation, there was no data available at FU 1, therefore, there is no grey bar at FU 1.

## **CONCLUSION**

This is the largest single study about outcome in CES after decompression. It displays unique data about the outcome of micturition, defecation, sexual function, sciatica and altered sensation of the saddle area as well as presenting features of a large cohort of CES patients, proving that micturition, defecation and sexual dysfunction are common both at presentation and at follow up. Female patients were found to have significant more defecation dysfunction at presentation than their male counterparts. A shorter time to decompression was a risk factor for sciatica shortly after surgery (at FU 1), but not for long term outcome. Studies on correlation between imaging at presentation and outcome could help identifying the patients being more at risk at presentation. Missing data were handled with multiple imputation with analysis of possible bias. A follow up study is recommended for more long term follow up data. In conclusion, recovery after decompression for CES does take a long time and is not complete in a substantial number of cases; something for which we should adequately prepare our patients when diagnosing CES.

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