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DOI:
10.1111/ene.14182

Document Version
Peer reviewed version

Link to publication record in King's Research Portal

Citation for published version (APA):
Gibson, L. L., Harborow, L., Nicholson, T., Bell, D., & David, A. S. (2020). Is scan-negative cauda equina syndrome a functional neurological disorder? A pilot study. *European Journal of Neurology*, *27*(7), 1336-1342. https://doi.org/10.1111/ene.14182

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Download date: 13. Jan. 2025

Is scan-negative cauda equina syndrome a functional neurological disorder? A pilot study.

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RUNNING TITLE: SCAN-NEGATIVE CAUDA EQUINA SYNDROME

Keywords: functional neurological disorders, cauda equina syndrome

Total word count: 3414

ABSTRACT

BACKGROUND
Cauda equina syndrome (CES) is a neurosurgical emergency which warrants a lumbar MRI. Many patients with suggestive symptoms of CES have no radiological correlate. A functional (non-organic) aetiology has been proposed in some, but currently little is known about this patient group and their clinical outcomes.

METHODS
At a tertiary referral centre, 155 adult patients underwent urgent lumbar MRI for suspected CES in one year from December 2014. Data regarding clinical symptoms and follow-up were obtained from records. Patients were divided into CES (n=25), radiculopathy (n=68) and scan-negative (SN) groups (n=62) from scans. Up to 3 years post-discharge, postal questionnaires were sent to patients with Owestry Disability Index, Pain Catastrophising score, PHQ-9, GAD-7, PHQ-15 and WSAS measures.

RESULTS
No clinical symptoms were found to differentiate CES from SN patients. Functional co-morbidities were significantly more common in SN patients but mental health diagnosis frequency did not differ. Follow-up was variable with no consistent referral pathways, particularly for the SN group. 33% (n=47) responded to postal questionnaires; high levels of pain, symptom chronicity and disability were ubiquitous but self-reported mental health diagnoses and PHQ-15 were higher for SN patients.

CONCLUSIONS
Conflicting data suggests further research is needed to investigate the prevalence of mental illness and somatic symptoms in SN cases. SN patients have higher rates of co-morbid functional disorders and inconsistent referral pathways. Self-report measures indicate impaired quality of life across all groups. The low response rate limits the generalisability of findings but neuropsychiatric assessment and care pathway optimisation should be considered.

INTRODUCTION

Cauda equina syndrome (CES) is a neurosurgical emergency caused by compression of the cauda equina nerve roots. The clinical presentation is variable but back pain with saddle anaesthesia, bladder, bowel or sexual dysfunction is highly suggestive and warrants an urgent lumbar MRI. A recent systematic review found 43-86% patients undergoing such scans have no structural pathology identified radiologically (1). These 'scan-negative' patients appear to be clinically indistinguishable from those with established CES, and no symptom, individually or in combination, has been found to reliably predict the presence of CES (2). The low correlation between symptoms and MRI necessitates scans in all patients (3-5). Currently little is known about this scan-negative patient group; questions remain regarding the cause of their symptoms, their clinical course on discharge from hospital and how best to treat them.

A functional aetiology or functional neurological disorder (FND), where impairment arises without structural abnormality, may underpin the disability for some (2, 6). While historically a psychological aetiology has been assumed to underpin FND, this has now largely been replaced by more nuanced biopsychosocial models (7). FND can cause almost any combination of neurological symptoms and as such can mimic almost any neurological disorder (8). Furthermore it is one of the commonest causes of neurological presentations, accounting for up to 30% of neurology outpatient consultations, such that it would be surprising if FND was not also encountered in neurosurgical settings like CES (9, 10). Indeed, in a prospective case series, 90% of scan-negative patients had a positive Hoover's sign, pointing to a functional aetiology, which was not present in any of the confirmed CES patients (6). Furthermore, the vulnerability to other functional disorders and higher rates of co-morbid mental health problems which are often seen with FND have also been found in scan-negative CES patients (11).

More detailed evaluation and longer follow-up are required to characterise this scan-negative group further, which would inform a range of clinical care pathways addressing their medical and psychiatric needs. We aimed to do this by collecting data retrospectively to include a period of follow up of 3 years to enable us to monitor the clinical evolution and impact on quality of life.

METHODS

This study received Health Research Authority approval from the West London Research Ethics Committee, the IRAS number is 222024. Informed consent was obtained from all patients completing a questionnaire in the follow-up study.

Patients. In one year from December 2014 to December 2015, 160 adult patients were identified from imaging records who underwent an urgent lumbar MRI for suspected CES in the emergency department at Kings College Hospital, a tertiary neurosurgery centre. 155 of these patients met the inclusion criteria with adequate clinical notes and no alternative cause identified accounting for their symptoms. Where more than one MRI was performed during this time scale, only the first was included.

Electronic records were retrospectively reviewed to identify demographic information, clinical symptoms, co-morbidities including functional and psychiatric diagnoses, 3 years of follow-up data and further MRI scans dating back to 2010. All available clinical notes, investigation requests and discharge summaries were used to collate these data. Data were collected regarding back pain, urinary symptoms: incontinence or retention (defined as $\geq$100ml post void residual volume), saddle anaesthesia, bowel incontinence or loss of anal tone, leg weakness, leg numbness and sciatica. As described above, five patients were subsequently excluded at this stage, two due to the insufficiency of their clinical records and three for whom, after reviewing their clinical notes or scan, it became apparent that an alternative diagnosis (multiple sclerosis in two and transverse myelitis for another) accounted for their presentation. *Figure 1.*

Postal surveys were sent to patients included in the follow up phase of the study except where they were repatriated to other local hospitals or self-discharged. Questionnaire measures included: the Owestry Disability Index (ODI), quantifying disability from back pain; PHQ-9, a standardised self-report depression screening questionnaire; GAD-7, a standardised self-report anxiety screening questionnaire, PHQ-15; a standardised screening instrument for somatisation; the Work and Social Adjustment Scale (WSAS), a measure of impairment in functioning; Pain Catastrophizing score and questions about follow-up and other co-morbidities. Several rounds of postal questionnaires were sent and telephone calls were also made to maximise rate of return. In a few cases, measures were completed over the telephone.

Definitions. CES was defined clinically as per guidelines proposed by Fraser et al. requiring as a minimum perianal sensory loss, bladder, bowel or sexual dysfunction (12). Radiologically CES was classified based on the MRI reports; if crowding or effacement of the thecal sac was noted the patient was considered to have cauda equina syndrome. Those without cauda equina compression were divided into radiculopathy, where nerve root impingement or contact was present, and scan-negative (SN). The radiculopathy group was considered separate as the root compression may have contributed to neurological symptoms whilst not being sufficient to account for the full CES picture.

Statistics. Chi-squared or Fisher's exact two-sided tests were used to compare categorical data; the CES group was compared with the radiculopathy and SN groups separately. For continuous data, one way ANOVA was used to compare respective groups. Statistics were performed using SPSS.

Figure 1. Flow chart of patient inclusion.

RESULTS

CES was confirmed radiologically in only 25 patients (16%). The remaining scans showed evidence of radiculopathy (n=68, 44%) or were SN (n=62, 40%).

Clinical presentation

SN patients were significantly younger than those with CES (mean age 45 vs 55, p = 0.005) and more likely to be female (77% vs 48%, p = 0.007). There was no individual symptom which was significantly more likely to occur in CES or SN groups. Bilateral sciatica and bilateral leg paraesthesia were significantly more common in patients with CES v radiculopathy (56% v 28%, p = 0.012; 28% v 7%, p = 0.014) but these were not distinguishing factors between CES and SN patients (56% v 37%, p =0.11; 28% v 24%, p = 0.74).

SN patients did present with significantly more functional co-morbidities, namely fibromyalgia, irritable bowel syndrome, dissociative seizures, chronic regional pain syndrome and non-cardiac chest pain than those with CES. There was no significant difference in frequency of reported mental health diagnoses documented between CES v radiculopathy or CES v SN groups. *Table 1.*

Follow up phase.

Follow-up over three years was reviewed for all 155 patients. No follow-up data were available for 12 patients who were repatriated to other local hospitals or one patient who self-discharged. Unsurprisingly, surgical intervention and neurosurgical follow up was significantly more common in the CES group versus the radiculopathy and SN groups, respectively (surgical intervention 84% vs 31%, and vs 3%, $p < 0.001$; neurosurgical follow-up 80% vs 38%, and vs 26%, $p < 0.001$).

Two SN patients received a surgical intervention despite there being no compromise of the cauda equina or nerve roots. In one case this was due to a historical lumbar fracture and in a second the clinical indication stated was excessive level of pain reported. In the CES group, 7 surgeries were performed electively outside the 48 hour recommended window. In two cases this was due to patient choice, another was due to a complicating deep vein thrombosis, one surgery was arranged electively at a nearby specialist centre and the remaining three were due to the treating team believing the clinical presentation at the time did not warrant immediate intervention, often on the basis of recent similar imaging studies.

Pain clinic, urology follow up and physiotherapy were generally equally spread between the groups, with the exception of significantly more CES patients receiving urological follow up than patients with radiculopathy (28% vs. 7%, $p = 0.03$). Those in the SN group were significantly more likely to receive either psychological input or no follow-up than those with CES (18% vs 0%, $p < 0.05$; 27% vs 0%, $p< 0.01$). *Table 2.*

Table 2. 3 year follow up for suspected CES.

Survey responses.

A third of all those contacted replied to the postal and telephone surveys (n=47, 33%). The response rate differed across the three groups with fewer patients in the radiculopathy group responding. The characteristics of those who did reply were similar to the overall group in terms of age and gender. High levels of self-reported pain and disability indexed by the ODI were seen across all groups (SN 49.2 v radiculopathy 29.8 v CES 36.4, $p = 0.14$). Pain catastrophizing scores were uniformly high. The symptoms in those with radiculopathy and SN were no more likely to resolve than in those with CES; less than 10% of SN patients

reported symptom resolution at 3 years follow-up. All groups showed similar but major impairment in work and social functioning.

Mild to moderate levels of anxiety and depression were self-reported in the GAD7 and PHQ9 in all groups. However, the frequency of self-reported mental health diagnoses was significantly different across the groups with SN patients reporting the highest frequency (77% vs radiculopathy 57% vs CES 30%, p = 0.04). In the PHQ 15, evaluating somatic symptoms, there were significant differences across the groups with SN patients reporting higher rates of somatisation (15.7 vs radiculopathy 11.9 vs CES 8.0, p = 0.01). *Table 3.*

Table 3. Mean scores on self-report postal questionnaires.

DISCUSSION

In this two-part study, consistent with previous studies, we found that individual clinical symptoms were unable to differentiate those with or without CES. Although not uniquely distinguishing, some demographic features such as younger age and female gender, were more commonly associated with SN patients, mirroring the demographics typical of FND where the gender bias can be as high as 3:1 (8). We did not find evidence of increased mental health diagnoses between the respective clinical groups recorded in case notes but functional co-morbidity was significantly more common in the SN group.

In those who returned the postal questionnaires, self-reported psychiatric comorbidity was significantly higher in SN patients vs. confirmed CES. The prevalence of self-reported mental health diagnoses in this group reached 77%, higher than both general population levels and those seen in studies of chronic back pain where a high level of psychiatric comorbidity is known to coexist (13). It is also markedly higher than the clinician reported prevalence across the whole SN group at admission, which was closer to 20%. Selection bias in those responding (and clinical under-detection) are likely to underpin this finding. Mental health diagnoses are under recorded in emergency departments (14) and a previous study identified high rates of mental health disorders (approaching 50%) in a similar population of scan-negative patients (11). Regardless of the precise cause, such elevated rates of self-reported mental illness highlight the high level of subjective distress in at least a subset of this patient group. Indeed, in our survey responses, we found that SN cases had equivalent

high levels of pain, disability, distress and symptom chronicity impairments to those cases with identified structural aetiology. We also found moderate levels of anxiety and depression, indicated by the PHQ9 and GAD7, in all groups, suggesting this may be secondary to symptoms, rather than aetiologically related.

To date, few studies have looked at long-term patient outcomes in either SN patients or post-surgically in CES. Studies within the CES population have primarily focussed on symptomatic outcomes looking at bladder, limb and bowel function with less than 10% of studies measuring the impact on quality of life (15). This is despite a recent international consensus paper highlighting mood and social functioning as some of the most important outcomes for patients with CES (16). Studies that have measured quality of life outcomes in CES illustrate high levels of chronic pain impacting physical and social functioning albeit with low sample sizes (17). Indeed, a recent assessment of the long-term post-surgical outcomes found that over a quarter of patients were unable to return to full employment with significantly poorer physical outcomes overall compared with the population average (18). This on-going symptomatic burden is reflected in our broad self-report measures illustrating a chronic impairment to quality of life across all groups, giving important insight into the persistent disability experienced by a proportion of post-surgical CES patients. Future studies should aim to characterise this further with a wider range of outcome measures not restricted to physical symptoms.

The heavy symptomatic burden in people with CES is not reflected in current clinical care pathways. This is particularly apparent for the SN group for whom there is currently no accepted treatment protocol, and in almost a third, no follow-up was received after discharge. Interestingly, the SN group had higher levels of co-existent somatic symptoms, mirroring the increased functional co-morbidity in this clinical group. This indicates a susceptibility to functional disorders, and it seems possible that for a subset of patients without identifiable structural aetiology, a FND underpins their chronic symptoms. Indeed, this supposition is supported by a large retrospective study where functional somatic and neurological disorders were more common in scan-negative patients than those with confirmed CES (11). Similarly, a smaller prospective study by the same group demonstrated positive clinical signs of functional leg weakness to be common in patients with a scan-negative symptomatic presentation (6). While work remains to be done to fully identify the primary aetiology, it does not seem that latent structural neurological disease is an underlying cause; we note only

three patients who were excluded due to alternative neurological diagnoses accounting for their clinical presentation in 3 years of follow up.

Limitations

There are a number of limitations which limit the scope of any conclusions we are able to draw. The initial data collection was dependent on routine clinician documentation which, due to the retrospective nature of the study, was not systematic and is vulnerable to missing data. This may explain the apparent absence of excess psychiatric comorbidity found previously in similar scan-negative groups (11). The completion rate for our questionnaire was low and likely to be biased despite over 3 rounds of mailings and telephone messaging, restricting the generalisability of our findings. There may be multiple reasons behind this including some wariness to a perceived 'psychological' emphasis of the study – although letters had the general hospital heading; disillusionment with the clinical service in some; or alternatively a relatively high spontaneous recovery rate in others. Nevertheless, those responses that we did receive give a clear insight into the degree of subjective distress experienced by a portion of this population and highlights a need for greater neuropsychiatric investigation and input.

Conclusions

Our work echoes earlier indications of a likely functional aetiology underpinning the clinical symptoms in at least a proportion of scan-negative CES patients (11). We demonstrate for the first time, high levels of somatisation, pain and symptomatic distress in this clinical group while simultaneously documenting the lack of adequate follow-up care. While further work needs to be done to elucidate the underlying causes of the disorder, it will also be important to focus on developing clinical care pathways to ensure that this group receives adequate and appropriate follow up in the future.

Disclosures of conflict of interest

None

Acknowledgments

Many thanks to FND Action who gave helpful feedback in the design of the study and questionnaires.

Funding

TN is funded by an NIHR Clinician Scientist Award. This Article presents independent research funded by the NIHR. The views expressed are those of the authors and not necessarily those of the NHS, the NIHR, or the Department of Health.
LG was awarded an Isaac Schapera grant to cover the costs of this research.

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