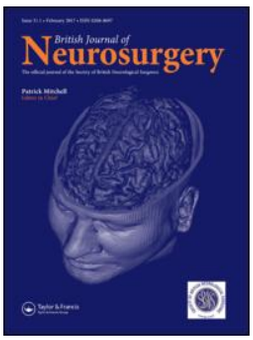

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ORIGINAL ARTICLE

Evaluation of nationwide referral pathways, investigation and treatment of suspected cauda equina syndrome in the United Kingdom

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ABSTRACT

Purpose: Cauda equina syndrome (CES) is a spinal emergency with clinical symptoms and signs that have low diagnostic accuracy. National guidelines in the United Kingdom (UK) state that all patients should undergo an MRI prior to referral to specialist spinal units and surgery should be performed at the earliest opportunity. We aimed to evaluate the current practice of investigating and treating suspected CES in the UK.

Materials and Methods: A retrospective, multicentre observational study of the investigation and management of patients with suspected CES was conducted across the UK, including all patients referred to a spinal unit over 6 months between 1st October 2016 and 31st March 2017.

Results: A total of 28 UK spinal units submitted data on 4441 referrals. Over half of referrals were made without any previous imaging ($n = 2572$, 57.9%). Of all referrals, 695 underwent surgical decompression (15.6%). The majority of referrals were made out-of-hours ($n = 2229/3517$, 63.4%). Patient location and pre-referral imaging were not associated with time intervals from symptom onset or presentation to decompression. Patients investigated outside of the spinal unit experienced longer time intervals from referral to undergoing the MRI scan.

Conclusions: This is the largest known study of the investigation and management of suspected CES. We found that the majority of referrals were made without adequate investigations. Most patients were referred out-of-hours and many were transferred for an MRI without subsequently requiring surgery. Adherence to guidelines would reduce the number of referrals to spinal services by 72% and reduce the number of patient transfers by 79%.

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KEYWORDS
Cauda equina syndrome; emergency surgery; spinal surgery; MRI; guidelines; referral

Introduction

Cauda equina syndrome (CES) occurs due to compression of the lumbosacral nerve roots that can lead to a constellation of symptoms including sphincter disturbance alongside lower limb motor and sensory deficits.[1] It is a common neurosurgical emergency with an incidence of approximately 0.3–0.5 per 100,000 per year.[2–4] Clinical features of CES have low sensitivity and specificity necessitating imaging early in the diagnostic pathway.[5–8]

Compounding these challenges is the current lack of consensus on how urgently decompressive surgery should be performed. There is no class I evidence to support emergency decompression at any time point. Meta-analyses have separately demonstrated statistically significant benefits of surgery within 24 hours,[9–11] within 48 hours,[12] and within 72 hours when treated as dichotomous variables.[11] Areas of contention leading to conflicting evidence include from what starting point the "time to surgery"

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Supplemental data for this article can be accessed here.

should be determined,[13] and if patients experiencing a complete injury with retention and overflow incontinence should be considered for emergency decompression.[9–11]

In the United Kingdom (UK) guidelines from the Society of British Neurological Surgeons (SBNS) and British Association of Spinal Surgeons (BASS) advise that patients presenting with acute back and/or leg pain with any bladder or bowel disturbance, with or without saddle sensory disturbance should be suspected of having CES. There should be a low threshold for investigation with emergency MRI at the hospital receiving the patient prior to referral to ensure timely diagnosis, referral and transfer to a specialist spinal unit where appropriate. Spinal units should not be considered a scanning service and out-of-hours MRI scanning should be considered routine practice to prevent needless and potentially harmful transport of patients for diagnostic imaging. If cauda equina compression is confirmed, guidance is that decompressive surgery be performed at the earliest opportunity.[14–17]

There is a paucity of literature regarding current service delivery against these standards. Consequently, we sought to investigate the current service provision for the diagnosis and management of CES across specialist units in the UK.

Materials and methods

A retrospective, multicentre observational study of the investigation and management of patients with suspected CES was conducted across neurosurgical units in the UK. Departments at each neurosurgical unit providing emergency spinal surgery (whether neurosurgery, dedicated spinal surgery, or orthopaedic surgery) were included and hereafter known as "spinal units". All patients with suspected CES referred within the six-month data collection period of 1st October 2016 to 31st March 2017 were included. The study protocol was approved by the audit and clinical governance committee of each participating hospital where required, the SBNS, and published on the website of the British Neurosurgical Trainee Research Collaborative (BNTRC).[18] Patient consent was not required due to the fully anonymised collection of data without any patient-identifiable information. The manuscript was prepared in accordance with the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines.

Data collection

Data were collected in each spinal unit using a standardised proforma by teams consisting of consultant surgeons, trainees, junior doctors, and medical students. Data were entered electronically into Castor EDC (Castor EDC, Amsterdam, Netherlands), which complies with all applicable laws and regulations (Supplementary Material Figure S1).

Table 1. Presenting symptoms of cauda equina syndrome as described in gleave and macfarlane.

CES Subgroup	Description
Incomplete (CESI)	Urinary difficulties of neurogenic origin, including altered urinary sensation, loss of desire to void, poor urinary stream and the need to strain in order to micturate.
With Retention (CESR)	Painless urinary retention and overflow incontinence, where the bladder is no longer under executive control.

Demographic data included the spinal unit, age, gender, source of referral and presentation categorised as incomplete CES (CESI) or CES with retention (CESR) (Table 1),[19] or "Other" for free text entry. Referrals for patients with isolated back pain or unilateral leg symptoms were subsequently excluded from calculation of referral timings. Imaging modality, imaging findings, imaging availability, purpose of referral, outcome of referral, patient transfers, surgical decompression, length of stay, and discharge destination were also recorded. Date and time information was collected for each step in the referral pathway (onset of symptoms, presentation to healthcare professional, referral to the spinal unit, MRI before or after referral, transfer, decompression and discharge including discharge destination). For the purposes of evaluating out-of-hours service provision, out-of-hours was defined as outside the hours of 9am–5pm Monday-Friday.

Statistical analysis

Categorical comparisons on pair-wise data were undertaken using Fisher's Exact testing. Continuous data excluding time intervals were analysed using Kruskal-Wallis testing. Time interval data were analysed using generalised linear modelling with logarithmic transformation of the timing variables following visual inspection of Q-Q plots. Univariable analyses were performed based on referral pathways. Multivariable analyses were performed including age, gender, presentation, timing of referral (dichotomised into in-hours and out-of-hours), pre-referral imaging and the referrer. Length of stay was calculated based on time from MRI to discharge if transferred to the spinal unit for an MRI, or time from decompression at the spinal unit to discharge. Variables evaluated for length of stay for those transferred for an MRI included age, gender, presentation, timing of referral, the referrer, whether or not the patient underwent decompression after the MRI, and discharge location. Cases with specified dates and times were used to analyse time intervals in hours, while all cases with dates submitted were included to analyse time intervals in days. Bonferroni correction was implemented to account for the multiple time points tested in the referral pathway, with a resultant threshold p-value of 0.007 (0.05/7) used to denote statistical significance. All statistical analyses were performed in R version 3.4.3 (R Foundation for Statistical Computing, Vienna, Austria).

Results

A total of 4441 referrals across 28 UK spinal units (coverage 93%) were included in the study during the six-month period. Median patient age at referral was 47 years (IQR 36–61). The majority of referrals were for female patients, and from a hospital other than the spinal unit (Table 2). Nearly half of the patients presented with CESI, and of those submitted as presenting with "other" symptoms, lower back pain was the most common (Supplementary Material Figure S2). Of all referrals, 3679 (82.8%) were made on a weekday. For referrals where time of referral were submitted, the majority were made out-of-hours ($n = 2229/3517$, 63.4%). This is in contrast to the time of presentation where available where less than half of patients presented out-of-hours ($n = 622/1261$, 49.3%) (Figure 1).

Referral and treatment characteristics

In total, 1628 referrals (36.6%) were made with an MRI completed prior to referral. Twenty percent of patients were referred

Table 2. Referral characteristics.

		All referrals ($n = 4441$)		Timing intervals ($n = 3168$)	
Group	Subgroup	*n*	%	*n*	%
Gender	Female	2654	59.8	1945	61.4
	Male	1784	40.2	1221	38.5
	N/A	3	0.1	2	0.1
Age	<16	11	0.2	11	0.3
	16–24	210	4.7	155	4.9
	25–49	2201	49.6	1606	50.7
	50–64	1034	23.3	745	23.5
	65–79	642	14.5	419	13.2
	80+	316	7.1	209	6.6
	N/A	27	0.6	23	0.7
Presentation	CESI	1979	44.6	1809	57.1
	CESR	860	19.4	772	24.3
	Other	1148	25.9	587	18.5
	N/A	454	10.2	0	0.0
Purpose of referral	MRI imaging and report confirming cauda equina compression	813	18.3	497	15.7
	MRI report only reporting cauda equina compression	105	2.4	37	1.2
	Requested advice whether to scan	1867	42.0	1349	42.6
	Requested to transfer for scan	783	17.6	700	22.1
	Requested review of MRI scan	355	8.0	225	7.1
	No compression of cauda equina on imaging but requesting neurosurgical management advice	293	6.6	194	6.1
	Requesting information how to proceed with investigation of ?CES	100	2.3	77	2.4
	Referred while waiting for MRI	75	1.7	47	1.5
	Other	50	1.1	42	1.3
Referrer	Other hospital	2485	56.0	1833	57.9
	Other specialty on same site as spinal unit	1194	26.9	867	27.3
	Primary care (GP)	715	16.1	431	13.6
	Other	41	0.9	32	1.0
	N/A	6	0.1	5	0.2
Imaging Pre-referral	None	2572	57.9	2008	63.3
	MRI	1628	36.7	964	30.4
	CT myelogram	3	0.1	2	0.1
	CT	116	2.6	94	3.0
	Plain film	89	2.0	74	2.3
	Other	27	0.6	23	0.7
	N/A	6	0.1	3	0.1

CESI: incomplete cauda equina syndrome; CESR: cauda equina syndrome with retention. N/A refers to missing data.

with reported MRI evidence of cauda equina compression ($n = 918/4441$, 20.7%). Of the 878 cases with an MRI report, the most common causes included disc prolapse ($n = 470/878$, 53.5%), spinal stenosis ($n = 205/878$, 23.3%) and spinal metastases ($n = 78/878$, 8.9%).

When referrals were made without an MRI, the purpose of the referral was for imaging advice ($n = 1564/2813$, 55.6%), the lack of availability of an MRI scanner out-of-hours ($n = 551/2813$, 19.6%) and the absence of an available MRI scanner to the referrer ($n = 265/2813$, 9.4%). Referrals from other hospitals were more likely to be made with a completed MRI ($n = 1033/2485$, 41.6%) than referrals from other specialties on the same site as the spinal unit ($n = 436/1194$, 36.5%, $p < 0.001$), or referrals from primary care ($n = 153/715$, 21.4%, $p < 0.001$) (Supplementary Material Figure S3 and Table S1).

Graphical depiction of the outcome of referrals is shown in Figure 2, demonstrating the pathways for patients with suspected cauda equina syndrome. Of the 2813 referrals made without an MRI, in 2336 cases the referral outcome was to perform an MRI scan (83.0%). In total, 695 patients referred underwent surgical decompression (15.6%). Of the patients referred with an MRI, 474/1628 (29.1%) underwent decompression. A significantly smaller proportion of referrals made without an MRI resulted in surgical decompression ($n = 221/2336$, 9.5%, $p < 0.001$). Causes were not significantly different between cohorts undergoing decompression with an MRI before or after referral which was performed most commonly for a disc prolapse ($n = 532/695$, 76.6%), spinal stenosis ($n = 96/695$, 13.8%), and infection (17/695, 2.4%).

Out-of-hours service provision in other hospitals

In other hospitals receiving patients with suspected cauda equina syndrome, the majority of referrals to the spinal unit were made out-of-hours ($n = 1529/2485$, 72.7%, Supplementary Material Table S1). Out-of-hours referrals were more likely to be made without a completed MRI scan (out-of-hours $n = 991/1529$, 64.8%, vs. in-hours $n = 202/575$, 35.1%, $p < 0.001$, Figure 3). Of the referrals made without an MRI scan, referrals out-of-hours were more likely to result in the transfer of the patient for an MRI scan (out-of-hours $n = 370/991$, 37.3%, vs. in-hours $n = 38/202$, 18.8%, $p < 0.001$). Overall 2.9% of all out-of-hours referrals from other hospitals underwent an MRI following referral which led to surgical decompression.

Referral timings

Time intervals were available for 3168 (71.3%) of referrals (Table 2) with analysis based on the pathways outlined in Figure 2. Comparisons were made for each stage in the referral process with each pathway relative to the most common (MRI not done, MRI at other hospital) (Figures 4 and 5). Full results including effect sizes and confidence intervals can be found in the Supplementary Material (Table S2).

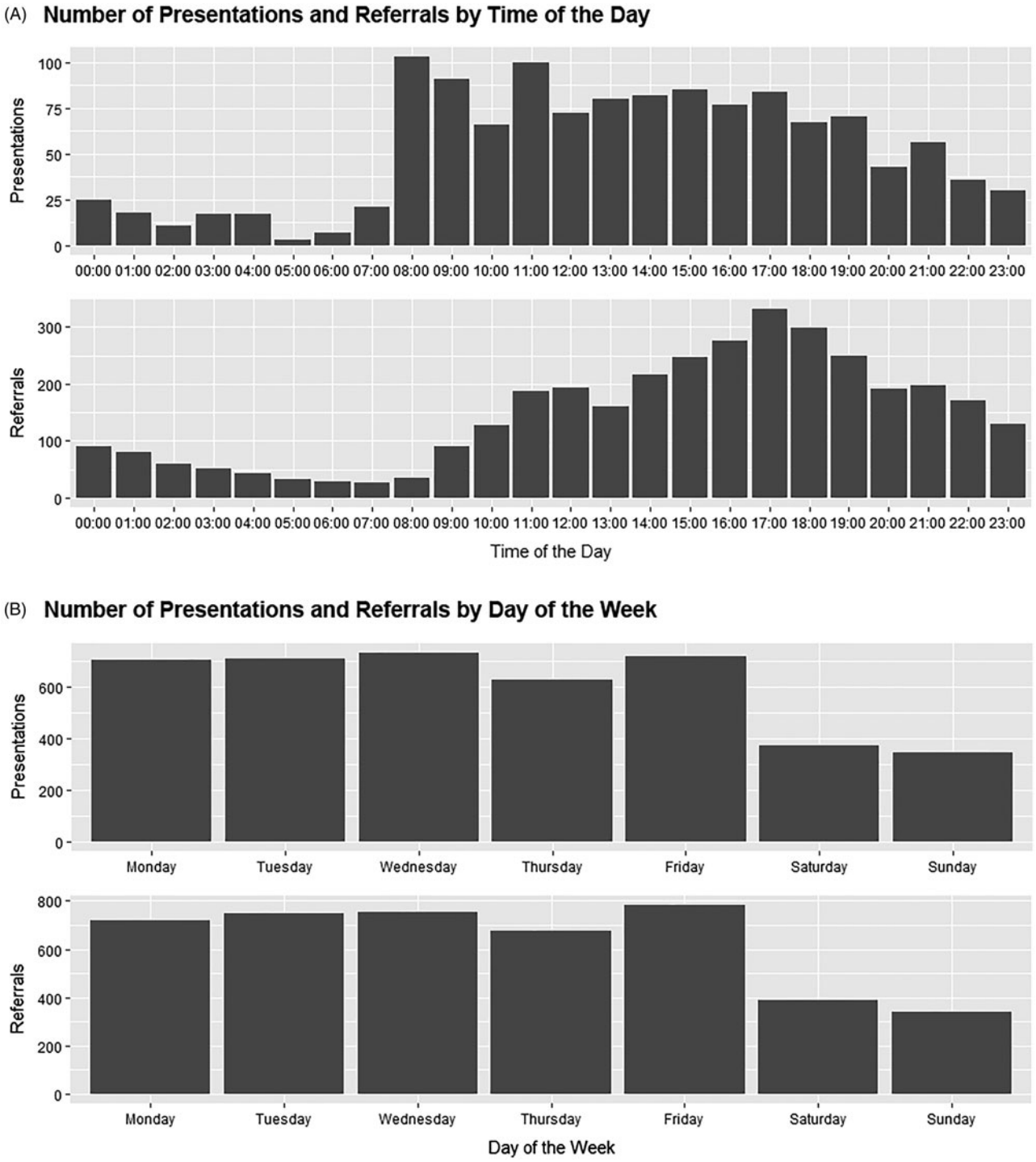

Figure 1. Referral Patterns for presentations and referrals for suspected Cauda Equina Syndrome (CES). A – Time of the day, B – Day of the week.

There was a significantly longer time interval from presentation to referral for patients undergoing an MRI prior to referral (median 6.1 vs. 1.5 hours, $p < 0.001$). Conversely, the time interval from presentation to MRI for patients referred with an MRI was significantly shorter than patients referred before an MRI (median 3.1 vs. 13.9 hours, $p < 0.001$). Of those cases referred before an MRI was completed, the time interval from referral to MRI was significantly shorter if the patient underwent the MRI at the spinal unit even if transferred from another hospital (median 7.2 vs. 13.3 hours, $p < 0.001$). Moreover, for patients referred from another hospital, the time interval from MRI to decompression was significantly longer in patients referred with an MRI compared to if referred before an MRI was completed (median 23.2 vs. 9.7 hours, $p = 0.003$). These results were consistent when repeating the analysis for time intervals expressed in days (Supplementary Material Table S3).

Full multivariable results are shown in Supplementary Table S4. Cases submitted with an MRI prior to referral reported a significantly longer time interval from presentation to referral and shorter time from presentation to MRI than cases referred without imaging. Referrals in-hours were associated with a shorter time from presentation to MRI, while patients presenting to primary care (GP) were referred earlier than patients from hospitals other than the spinal unit. Cases referred from the hospital in

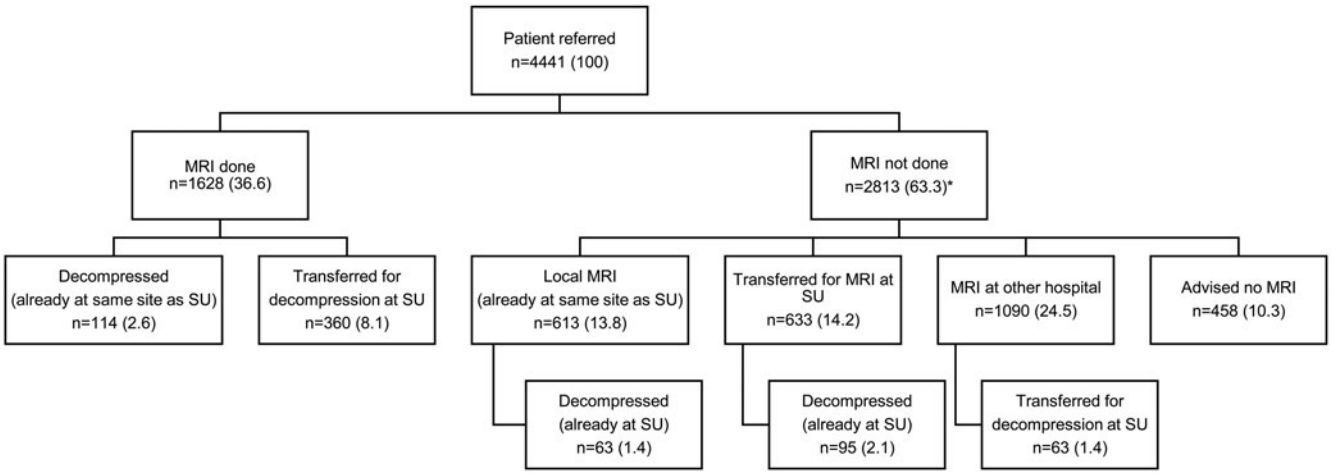

Figure 2. Referral Outcome for Patients with suspected Cauda Equina Syndrome (CES). Numbers shown with percentages in brackets of the total sample ($n = 4441$). SU: Spinal Unit. *Nineteen cases were submitted with no MRI completed and no referral decision made.

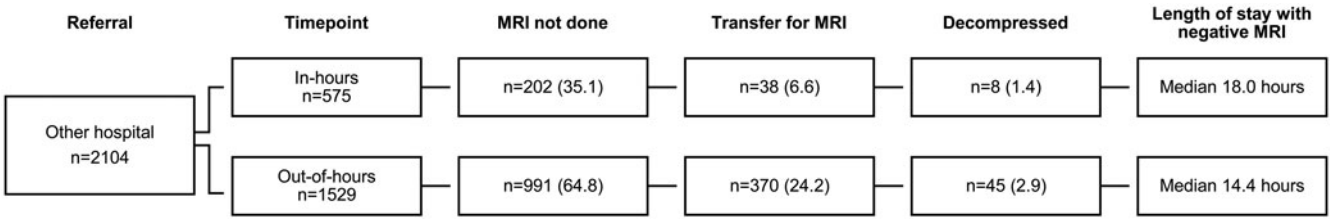

Figure 3. Comparison of in-hours and out-of-hours referrals from hospitals other than the spinal unit. Percentages shown refer to the total number of referrals by timepoint.

the same site as the spinal unit reported a significantly shorter time interval from presentation to MRI and presentation to decompression. Increasing age was associated with a longer time interval from MRI to decompression and referral to decompression. These results were consistent when analysing time intervals expressed in days (Supplementary Material Table S5).

Length of stay

Median length of stay for patients transferred for an MRI was 17.9 (IQR 4.5–70.3) hours. Median length of stay for patients transferred not requiring surgery was 11.1 (IQR 3.6–48.6) hours compared to 75.6 (41.9–116.5) hours for those requiring decompressive surgery. A quarter of patients transferred for an MRI scan not requiring surgery were admitted for over 24 hours ($n = 78/330$, 23.6%). Length of stay was significantly shorter if patients not requiring surgical decompression were transferred back to their referring provider (median 7.2 (IQR 3.0–15.4) hours) rather than their original place of residence (median 16.8 (IQR 4.0–59.9) hours, $p < 0.001$). For all patients undergoing surgical decompression, multivariable analysis demonstrated a longer admission was associated with increasing age (Wald $Z = 2.94$, $p = 0.003$), and diagnosis of an infection (Wald $Z = 2.86$, $p = 0.004$) or spinal stenosis (Wald $Z = 3.03$, $p = 0.003$). A shorter admission was associated with referrals from primary care compared to other hospitals (Wald $Z = -2.35$, $p = 0.019$, Supplementary Material Table S6–S7).

Discussion

In this retrospective multi-centre study of 4441 referrals for suspected CES, substantial deviations from UK guidelines were identified. While the SBNS and BASS recommend that local hospitals should investigate patients thoroughly prior to referral to spinal services, in this study only a minority of cases were referred with diagnostic imaging completed ($n = 1628/4441$). Guidelines also state that spinal units should not be considered a scanning service, but due to the majority of referrals being made out-of-hours ($n = 1529/2104$, 72.7%) a proportion of patients were transferred to the spinal unit for an MRI scan ($n = 370/1529$, 24.2%). Importantly, of these referrals a fraction required surgical decompression ($n = 45/1529$, 2.9%).

Referral pathway

Referrals made without an MRI were most commonly made for advice on whether an MRI was indicated. Existing guidelines already include specific signs and symptoms suggestive of CES available for all practising physicians that should prompt an emergency MRI.[20,21] A proposed diagram based on these guidelines is shown in Figure 6, including where the MRI should be performed and what MRI findings require an emergency referral to spinal services. Based on a six-month period, implementing this pathway would reduce the number of referrals to spinal services by 72% (4441 to 1224) and reduce the number of patient transfers by 79% (739 to 156). This pathway should be incorporated into local and regional protocols with dissemination in key departments particularly Emergency Departments. Due to the possibility of self-referral, direct-to-patient education leveraging existing organisations such as Cauda Equina UK could also be made to improve detection of red flag symptoms early on in the disease.[22] Given that the majority of patients presented in-hours and were only referred out-of-hours, work to improve triage of these patients will also improve the burden of undertaking diagnostic imaging for these patients out-of-hours.

In other countries, guidelines are less specific on when and where an MRI should be undertaken. In the Netherlands,

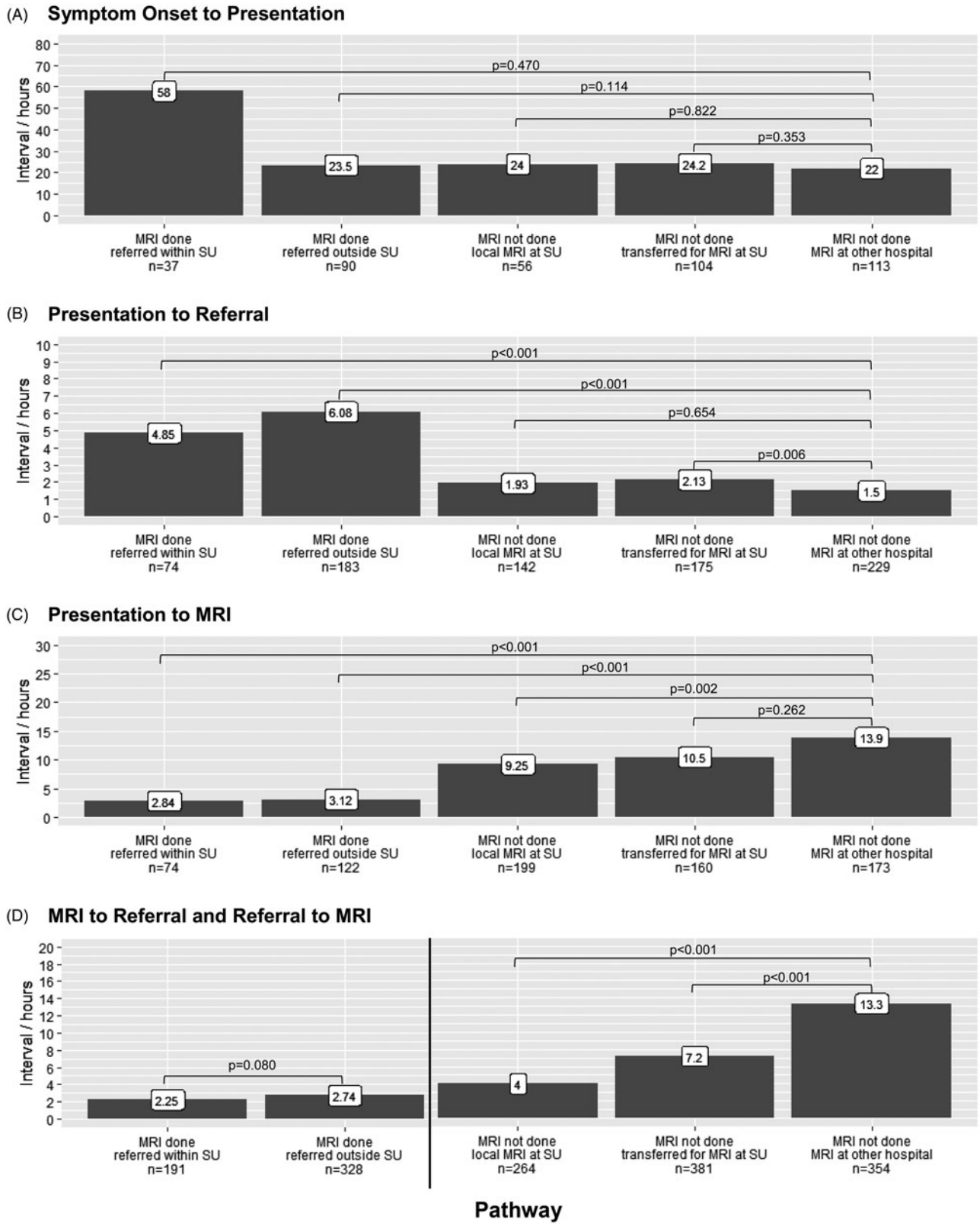

Figure 4. Breakdown of time intervals in median hours per stage in the referral process, with p values relative to the reference pathway (MRI not done, MRI at hospital other than the spinal unit). SU – Spinal Unit. A – Symptom onset to presentation, B – Presentation to referral, C – Presentation to MRI, D – MRI to referral and referral to MRI.

referral to specialists should be considered when the general practitioner is not sure about the diagnosis or considers surgical intervention.[23] In Norway and Denmark, guidelines for degenerative spinal conditions recommend referral to neurosurgery after an MRI is completed, but in the case of suspected CES early referral is recommended without specific guidance on where diagnostic imaging should be performed.[24,25] In Germany, acute inpatient admission is required

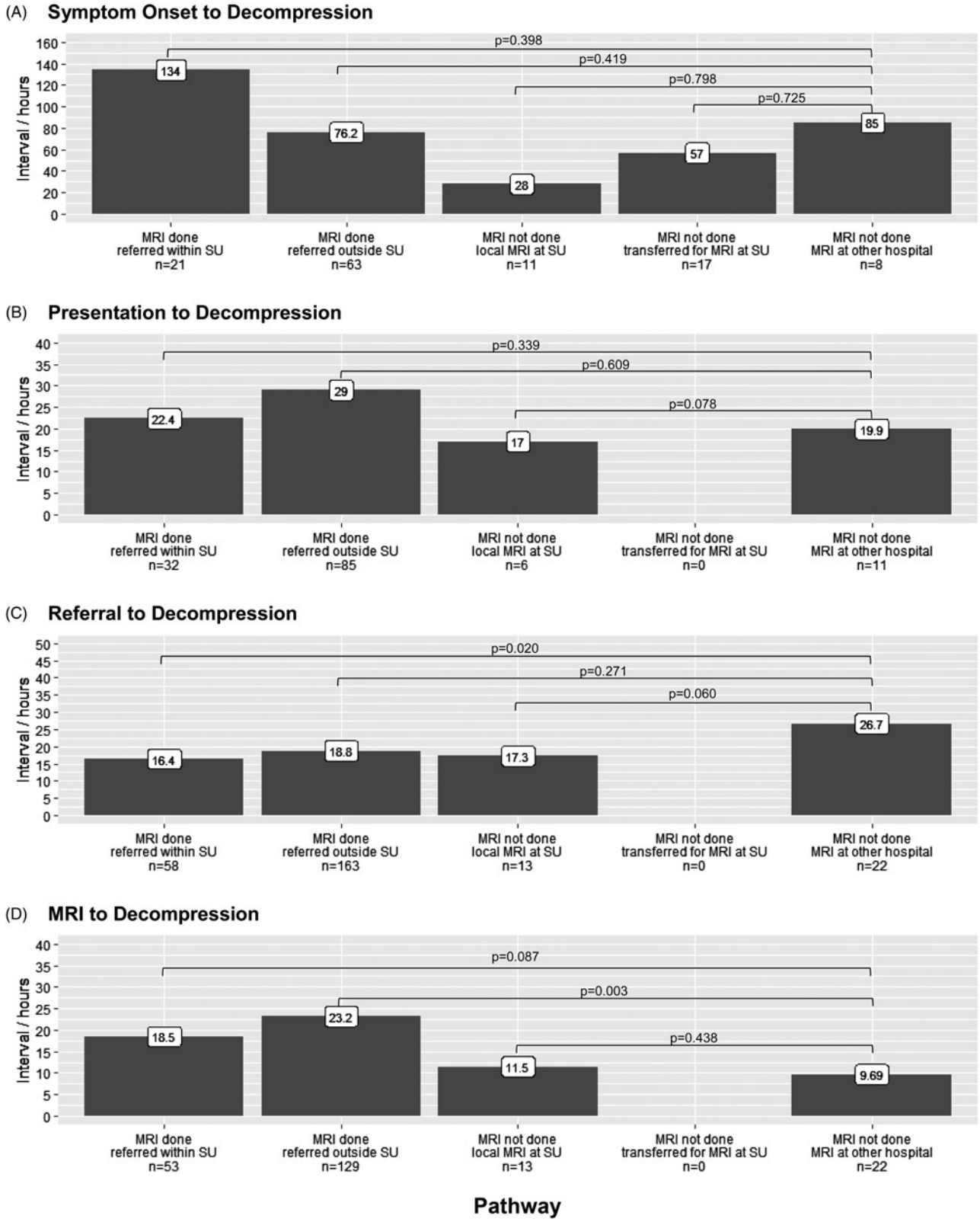

Figure 5. Breakdown of time intervals in median hours from each stage in the referral process to decompression, with p values relative to the reference pathway (MRI not done, MRI at hospital other than the spinal unit). SU: Spinal Unit. A – Symptom onset to decompression, B – Presentation to decompression, C – Referral to decompression, D – MRI to decompression.

in patients with "red-flag" symptoms but again no specific guidance on location for diagnostic imaging is given.[26] Overall, no study of referral patterns or pathways for suspected CES in other countries was identified.

Diagnostic imaging availability

Part of the problem with implementing the pathway outlined in Figure 6 lies in the availability of diagnostic imaging. Numerous national guidelines and protocols recommend that emergency

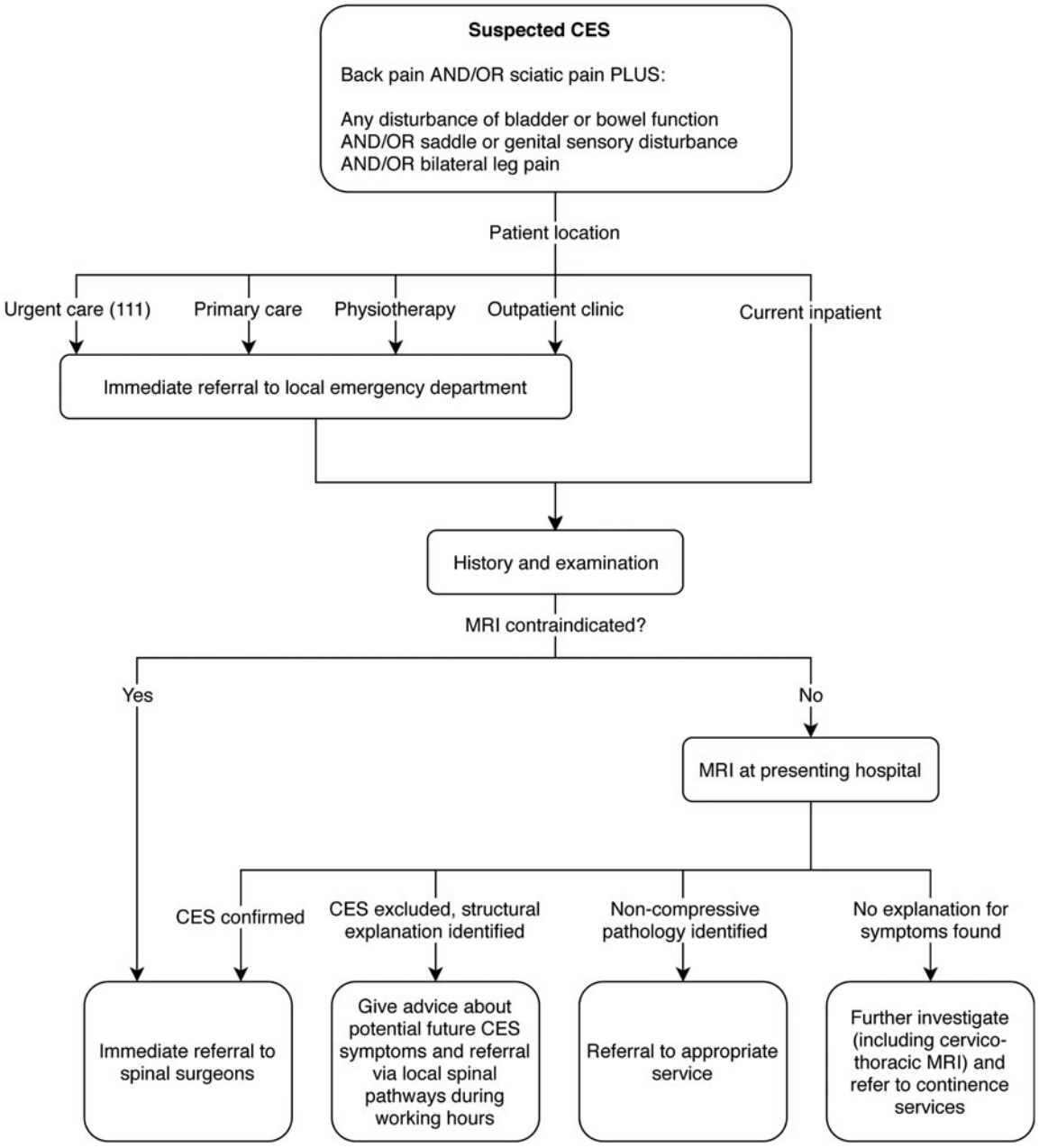

Figure 6. Proposed Flow Diagram for Referrals for Suspected Cauda Equina Syndrome (CES) in the United Kingdom.

MRI scanning is available 24/7 in all UK district general hospitals[20,27,28] and yet UK survey data showed only 14% of hospitals providing access to MRI 24 hours a day.[29] Although costs and staffing levels are often cited as the challenge, there are examples of cost neutral solutions without increased staffing, such as arranging out-of-hours CT radiographer training in basic brain and spine MRI scanning with rotations through MRI one week in every twelve.[29]

Any associated cost increase will have to be balanced against two current significant cost burdens. First, the costs associated with the transfer of patients for an MRI scan which one hospital estimated at £6,000 per referral. Such is the cost that if only two referrals were made from one district general hospital it would be more cost effective for the trust to have their own dedicated radiographer on call.[30] Second, the clinical and financial implications of inadequate investigation and management of CES are an important consideration.[31] Around 10% of CES cases involve litigation.[1] Average claims range from £117,331, to £211,758 per case and a highest settlement of £2,041,000.[32]

Timing of surgery

Although the decision to perform decompressive surgery for confirmed CES is a recognised emergency, the decision as to how quickly this should be performed is unclear. Guidelines are based on balancing the duration and clinical course of symptoms and signs alongside potentially greater risks of operating out-of-hours. No specific standards on time intervals have been published due to the absence of consistent results in published studies. The literature is also unclear with regards to the defined starting points used to calculate "time to surgery", with studies reporting the time from the first urinary symptom, time from bladder paralysis, or the time from admission to hospital.[13]

This study revealed no significant differences between referral pathways and the time of onset of symptoms or presentation to decompression. However, pathways of patients referred outside the spinal unit were associated with significantly longer time intervals from referral to MRI if undergoing the MRI outside the spinal unit after referral. Furthermore, time intervals from MRI to decompression were significantly longer for patients with a confirmed CES undergoing an MRI before referral. Both represent the proposed pathway for managing patients with suspected CES in the UK. These results therefore have important implications for service delivery, particularly with efforts to increase the proportion of patients investigated through this pathway. Any policy changes to reduce the referral burden on specialist spinal units will need to ensure this does not result in an increase in the length of time patients with suspected cauda equina syndrome are being investigated and treated.

Future directions

The comparative epidemiology and referral patterns between countries for suspected CES requires further investigation. The Getting It Right First Time (GIRFT) is a programme delivered in the United Kingdom to improve the quality of care within the National Health Service by reducing unwarranted variations. Data from this study will inform work to review local policies related to out-of-hours arrangements for radiography to ensure compliance with national guidelines and delivering of 24-hour local MRI scanning.[30] This study has also informed an ongoing large prospective study of the investigation, management and outcome of patients with confirmed CES in the UK (Understanding Cauda Equina Syndrome or UCES) where comprehensive presenting symptomatology, timings, and outcome data will be collected.[33] The results of UCES will contribute to existing work developing tools to improve the clinical assessment and investigation of patients with suspected CES.[20,34]

Limitations

This study covered a large number of referrals to spinal centres within the UK over the six-month study period and 28 out of the 30 UK neurosurgical centres participated. However, the absence of two neurosurgical centres and smaller orthopaedic centres that did not participate, means that this does not cover the whole population of the UK. Despite these limitations, we think that the large number of cases in this study should be representative of those referred across the UK. There may be a variability in data collection between centres which could not be centrally verified by the primary investigators due to the anonymous nature of data collection. This study only addressed cases which were referred to a spinal unit, missing patients who were managed without an MRI or who underwent MRI scanning locally which was normal without referral to the spinal unit. Outcome data was not collected in this study; the aforementioned future prospective study of confirmed CES in the UK (UCES) will collect outcome data and its relationship to presenting symptoms and the referral pathway including timings will be identified.[33]

Conclusion

Guidelines in the UK for the investigation and management of patients with suspected cauda equina syndrome emphasise the importance of thorough investigation in the receiving hospital prior to referral to specialist units. This national retrospective multi-centre study of 4441 referrals has identified deviation from national guidelines. Currently the vast majority of referrals for suspected CES are made without an MRI out-of-hours with specialists subsequently recommending an emergency MRI that, in a proportion, requires a transfer to the spinal unit. Adherence to guidelines would reduce the number of referrals to spinal services by 72% (4441 to 1224) and reduce the number of patient transfers by 79% (739 to 156). Changes required to adhere to guidelines will need to acknowledge the identified longer time intervals for diagnostic imaging if patients are investigated locally without transfer to ensure the best care for patients with suspected CES in the future.

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