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ORIGINAL ARTICLE

The international standards for neurological classification of spinal cord injury: relationship between S4-5 dermatome testing and anorectal testing

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Study design: Prospective cross-sectional multicenter study.
Objective: To evaluate the correlation, sensitivity, specificity and predictive values of S4-5 dermatome and the anorectal examination for determination of sacral sparing in the International Standards for Neurological Classification of Spinal Cord Injury (ISNCSCI) examination.
Setting: Two tertiary hospitals that specialize in pediatric spinal cord injuries.
Methods: In all, 189 patients who were at minimum 3 month after spinal cord injury participated in complete ISNCSCI examinations. All examiners completed training for the proper completion of the ISNCSCI examination. Correlations and sensitivity/specificity analyses were conducted between S4-5 dermatome testing and the anorectal examination. Results were analyzed by age of patient, examiner, tetraplegia/paraplegia classification and injury level (T10-S3, L1-S3 and S3).
Results: The correlation between S4-5 dermatome and anorectal sensation was moderate (0.62, $P<0.001$). Using the anorectal examination as the gold standard, the sensitivity of S4-5 testing was 0.60 (0.49, 70) and specificity was 0.96 (0.90, 0.99). No single age group, tester, level, or type of injury differed from the overall result.
Conclusion: In the pediatric population, the correlation between S4-5 and anorectal sensation was lower than anticipated. The sensitivity of 0.62 for S4-5 testing and diminished sensation between T10 and S3 suggests that anorectal testing may either be a more sensitive representation of S4-5 function or activate an alternative neuronal pathway that is perceived by the patient. Further investigation into the validity of the sacral sparing components of the ISNCSCI examination is warranted.
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Keywords: spinal cord injury; ISNCSCI; outcome; sacral sparing; ASIA; anorectal examination

Introduction

The International Standards for Neurological Classification of Spinal Cord Injury (ISNCSCI) is the gold standard for evaluating and classifying the neurological consequence of spinal cord injury (SCI).[1] The examination includes three main sections, sensory, motor and anorectal testing. A completed examination provides a sensory score, sensory level, motor score, motor level, a neurological level and sacral sparing.

Sacral sparing is defined as volitional control and/or sensory preservation of the lowest segment of the cord (S4-5). Evaluation of sacral sparing also includes three components (1) sensory testing for appreciation of discriminatory ability (sharp\dull via using a sterile safety pin prick (PP) and of light touch (LT) perception at the musculocutaneous junction (2) testing of deep anal pressure during an internal rectal examination by applying pressure to the rectal wall and (3) testing for the presence of volitional anal contraction during the internal rectal examination. If sensation or volitional contraction is present, the SCI is defined as incomplete. As per the 2002 ISNCSCI manual, deep anal sensation (AS) testing is required because its presence 'can occasionally be the only evidence of a clinically incomplete SCI.' When classifying the severity of a SCI, absence of sacral sparing is considered as an complete injury, American Spinal Injury Association Impairment Scale A (AIS A, ISNCSCI manual p.11).[1]

The ISNCSCI manual instructions for testing and interpretation of deep anal pressure and contraction are vague.[1] In our experience, youths of any age had difficulty understanding the test instructions (for example, 'squeeze as if to

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hold in a bowel movement') if their injury occurred before bladder and bowel continence.[2,3] Additionally, there are no recommendations in the manual for discerning if the perception of sensation is a reaction to actually feeling the stimulus or as a result of triggering alternative pathways that result in other forms of sensation, such as warmth or tingling in the head, goose bumps or tingling in the legs. To compensate for the lack of manual recommendation and in an attempt to minimize false-positives, we standardized a protocol for the anorectal examinations.[3] Despite this standardized protocol for anorectal examinations, the test is difficult to perform in young children and, for pre-teenagers and adolescents, modesty becomes a factor that can potentially influence the accuracy of the test results. An alternative to the internal anorectal examination would be useful and it would be valuable to better understand whether S4-5 dermatome scores could be the sole source for determining AIS classification.

Towards this aim and as part of a larger, multicenter study on the ISNCSCI, we explored the relationship between the results of the internal rectal examination of deep anal pressure/contraction and results of S4-5 dermatome testing.[2–5]

Materials and methods

This study is part of a larger trial looking at the reliability of the ISNCSCI examination in children and youths with SCI.[2,3,5] This multicentered study was reviewed and approved by the Institutional Review Boards at participating sites. Written informed consent was obtained from parents or legal guardians of all subjects under the age of 18 years old and participants between ages of 7 and 18 completed informed assents.

This study included the analysis of the first of four repeated ISNCSCI exams of participants enrolled in the larger reliability study. In total there were 189 subjects between the ages of 6 and 21 with chronic SCI ($\geqslant$3 months after injury) that were included in the analysis (Table 1). The distribution of AIS classification was: 49% AIS A, 24% AIS B, 15% AIS C and 11% AIS D. The average age of the study population was 14.6 ± 4.2 years with an average of 5.0 ± 4.4 years since injury.

The ISNCSCI examination was completed using the ISNCSCI testing protocol described in the manual published by the American Spinal Injury Association.[1] AS was tested by having the examiner gently applying pressure to the rectal wall a minimum of three times. If, without prompting, the subject accurately identified examiner applying pressure, they were scored as having rectal sensation. If the subject's response was inconsistent, a minimum of 8 out of 10 accurate responses was required for designating the injury as incomplete. After sensation testing, the subject was asked to squeeze 'as if to hold in a bowel movement'. Special attention was taken to ensure the participant did not increase intra-abdominal pressure by holding their breath or tighten their abdominal muscle.

As defined by the ISNCSCI manual, sacral sparing defined as any sensation at S4-5 (either LT or PP), AS and/or anal contraction. For S4-5 dermatome, scores were combined to create a binary outcome, with 'negative' defined as no S4-5 LT/PP right/left side sensation, and 'positive' defined as the presence of any sensation at S4-5 (LT/PP, right or left side). All examiners were trained in the examination as described by the American Spinal Injury Association manual by an expert in the conducting and scoring ISNCSCI examination.[6]

The ISNCSCI manual states that 'the presence of deep AS can occasionally be the only evidence of a clinically incomplete SCI'.[1] This implies that deep AS is the definitive test for sacral sparing. For that reason, sensitivity/specificity analysis was completed using deep anal pressure as the gold standard for sacral sparing and S4-5 sensation as a test of sacral sparing. To evaluate the completeness of neurological injury, patients were classified as 'positive' if they had any sensation between dermatome areas T10-S3, L1-S3, or at S3. If no sensation was present in those ranges, they were classified as 'negative'.

Data were analyzed using SAS V9.1 statistical software (SAS Institute, Cary, NC, USA). Spearman correlations were used to examine the relationship between S4-5 dermatomes, AS and anal contraction. Sensitivity, specificity, receiver operating characteristics area under the curve (ROC AUC) and predictive values were calculated along with the 95% confidence intervals as measures of test accuracy. Sensitivity analysis for the influence of each tester was completed by re-running the analysis after the removal of each tester.

We certify that all applicable institutional and governmental regulations concerning the ethical use of human volunteers/animals were followed during the course of this research.

Table 1 General demographics of sample population

	Years	
Average time since injury	5.0 ± 4.4	
Average age	14.6 ± 4.2	
	Number	*Percent*
Gender		
Male	110	58
Female	79	42
Severity		
Complete	93	49
Incomplete	96	51
Type of Injury		
Tetra	98	52
Para	91	48
ASIA impairment scale		
ASIA A	93	49
ASIA B	45	24
ASIA C	28	15
ASIA D	21	11
Not scorable	2	1

Abbreviation: ASIA, American Spinal Injury Association.

Results

Statistically significant correlations were found between the presence of any S4-5 dermatome sensation (that is, LT/PP,

right or left with a score of 1 or more) with deep anal pressure ($r=0.62$, $P<0.001$) and anal contraction ($r=0.45$, $P<0.001$).

Of the 189 participants in this study, 96 did not have preservation of deep anal pressure (Table 2). Within this group, 3 (3%) tested positive for any S4-5 sensation (false positive) and 93 (97%) did not have any sensation with S-4 (true negative). In all, 93 tested positive for deep anal pressure. Of those, 56 (60%) had some sensation at S4-5 (true positive).

Using deep anal pressure as the gold standard of sacral sparing, the sensitivity of S-45 testing to determine deep anal pressure was 0.602 (95% CI 0.495, 0.771) and the specificity was 0.969 (95% CI 0.905, 0.992, Table 3). The positive predictive value was 0.949 (95% CI 0.849, 0987) and the negative predictive value was 0.715 (95% CI 0.629, 0.789). ROC AUC was 0.79 (95% CI 0.73, 0.84)

By age group, sensitivity was similar for age groups 6–11 (0.63; 95% CI 0.39, 0.83) and 16–21 (0.63; 95% CI 0.48, 0.76) years old. For 12–15 years old sensitivity was lower at 0.52 (95% CI 0.32, 0.72). For negative predictive value the largest difference was between patients between ages 12 and 15 (0.66; 95% CI 0.48, 0.80) and 6 and 11 (0.78; 95% CI 0.60, 090) year olds. Specificity and positive predictive value did not differ by age and were above 0.93 for all ages. ROC AUC was greatest for ages 6–11 (0.83; 95% CI 0.70, 0.73) years olds, followed by 16–21 (0.79; 95% CI 0.72, 0.87) years and then 12–15 (0.74; 95% CI 0.63, 0.85) year olds.

Patients with paraplegia had lower sensitivity (0.49; 95% CI 0.32, 0.06 versus 0.67; 95% CI 0.54, 0.70) but higher negative predictive value (0.75; 95% CI 0.64, 0.084 versus 0.066; 95% CI 0.48, 0.80) and those with tetraplegia. For both tetraplegia and paraplegia Specificity and positive predictive value was above 0.93. ROC AUC was greater for patients with tetraplegia (0.81; 95% CI 0.74, 0.88) compared with those with paraplegia (0.73; 95% CI 0.65, 0.82).

Sensitivity analysis using ROC AUC revealed that no single tester differed from the others or unduly influenced the results.

Table 2 2×2 contingency table comparing presence of deep anal pressure and any sensation at S4-5 dermatome

	No deep pressure	*Presence of deep pressure*	*Total*
Presence of S4-5 sensation	3 (3%)	56 (60%)	59
No S4-5 sensation	93 (97%)	37 (40%)	130
Total	96	93	189

Discussion

This is the first study to evaluate the correlation between AS and S4-5 dermatome sensation when testing for sensory completeness in children and adolescents with SCI. The AIS manual suggests, 'the presence of deep AS can occasionally be the only evidence of a clinically incomplete SCI' (p.11). It is essential clinicians do not assume the absence of S4-5 dermatome sensation equates injury completeness. Using AS as the definitive determinate of sacral sparing, the presence of S4-5 sensation demonstrated poor-moderate test sensitivity (0.60). If these two components of the ISNCSCI were assessing the similar cord segments, a higher sensitivity and correlation would have been expected.

In our study population of 93 subjects with deep anal pressure, 37 (40%) had no S4-5 dermatome sensation. The poor sensitivity suggests that using S4-5 alone would yield a high number of false negatives. The high test specificity (0.96) of S4-5, suggests that the absence of S4-5 sensation was a good predictor of the absence of AS.

Recognizing the difficulty that younger children may have with the examination, patients were stratified by age groups. The test sensitivity remained equally lower in 6–11 year olds compared with the 16–21 year olds suggesting the test is not influenced by age (Table 3). As this latter group could be considered an adult population, further exploration of the sacral sparing test components is warranted in adults.

Table 3 Sensitivity, Specificity, positive predictive value and negative predictive value of S4-5 dermatome testing with deep anal pressure

		Sensitivity (95% CI)	*Specificity (95% CI)*	*PPV (95% CI)*	*NPV (95% CI)*	*ROC AUC (95% CI)*
All subjects	189	0.60 (0.50, 0.70)	0.97 (0.90, 0.99)	0.94 (0.85, 0.99)	0.72 (0.63, 0.79)	0.79 (0.73, 0.84)
Age groups (years)						
6–11	44	0.63 (0.39, 0.83)	1.00 (0.83, 1.00)	1.00 (0.70, 1.00)	0.78 (0.60, 0.90)	0.82 (070, 0.73)
12–15	49	0.52 (0.32, 0.72)	0.96 (0.77,1.00)	0.93 (0.64, 1.00)	0.66 (0.48, 0.80)	0. 74 (0.63, 0.85)
16–21	93	0.63 (0.48, 0.76)	0.95 (0.83, 0.99)	0.94 (0.78, 0.99)	0.70 (0.57, 0.81)	0.79 (0.72, 0.87)
Type of injury						
Tetraplegia	98	0.67 (0.54, 0.70)	0.95 (0.82, 0.99)	0.93 (0.64, 1.00)	0.66 (0.48, 0.80)	0.81 (0.74, 0.88)
Paraplegia	91	0.49 (0.32, 0.66)	0.98 (0.89, 0.99)	0.94 (0.71, 1.00)	0.75 (0.64, 0.84)	0.73 (0.65, 0.82)

Abbreviations: CI, confidence interval; NPV, negative predictive value; PPV, positive predictive value; ROC AUC, receiver operating characteristics area under the curve.
Sensitivity = conditional probability that the test will be positive if the condition is present).
Specificity = conditional probability that the test will be negative if the condition is absent.
Positive predictive value = conditional probability that the condition is present if the test is positive.
Negative predictive value = conditional probability that the condition is absent if the test is negative.
ROC AUC = combination of sensitivity and specificity with values closest to 1.0 indicating highest quality of a diagnostic test.

Table 4 Description of sensation sparing from dermatomes T10-S3, L1-S3 and S3

		False negatives			*True positives*			*True negative*			*False positives*		
	Total	*#*	*Within group*	*Of all FN*	*#*	*Within group*	*Of all TP*	*#*	*Within group*	*Of all TP*	*#*	*Within group*	*Of all TP*
Total	189	37	20%		56	30%		93	49%		3	2%	
Sensation													
T10-S3	105	20	19%	54%	54	51%	96%	28	27%	30%	3	3%	100%
L1-S3	76	14	18%	38%	54	71%	96%	5	7%	5%	3	4%	100%
S3	53	1	2%	3%	52	98%	93%	0	0%	0%	2	4%	67%

Abbreviations: FN, false negative; TP, tetraplegia.

Looking by the type of injury, there was lower test sensitivity for patients with paraplegia compared with those with tetraplegia. There is no clear explanation for the low test sensitivity in patient with paraplegia. Previous work by Vogel *et al*.[3] demonstrated lower intratester reliability for patient with paraplegia, particularly in children between 6 and 15 years old. It is possible that the discordance in S4-5 dermatome with AS testing in patient with paraplegia is a reflection of tester reliability.

A striking finding of this study was the lack of continuity of dermatome sensation in patients who were classified as false negatives. In this subgroup of 37 patients, the presence of sensation between the levels T10-S3, L1-S3 and S3 progressively decreased from 54, 38, to 19%, respectively (Table 4). This is in contrast to the 56 subjects who were tetraplegia in which, presence of any sensation between T10-S3, L1-S3 and S3 was 96, 96 and 93%, respectively. The presence of sensation in tetraplegias suggests a zone of partial preservation spanning the neurology level of injury to the end of the cord, as would be expected in an incomplete injury.

The cause for the low test sensitivity and lack of zone of partial preservation in false negatives patients is unclear. Similar to other studies, the results of our study question the utility of sacral sparing as defined by the manual.[3,7] As already discussed, Vogel *et al*. found poor reproducibility of the anorectal examination in the majority of subjects with tetraplegia or paraplegia. Van Middendorp *et al*.[8] found that anal contraction, PP and LT, but not AS was predictive of walking in the acutely injured patients. They suggested the presence of AS might not be specific to an incomplete injury and hence future ambulation.

There is mounting evidence that AS might be perceived by patients through an alternative pathway. In 2004, Finnerup *et al*.[9] described a subset of SCI patients who were clinically tested as complete injuries, but could perceive sensation well below there injury. Using a term coined in the late 1990s, the authors described these patients as 'discomplete'.[9–11] In a more recent study Wietek *et al*. used functional MRI to determine whether anorectal stimuli would elicit cortical activation measured by functional magnetic resonance in clinical complete SCI. In their case series of 11 patients, 4 experienced sensation of gas or stool pressure in their middle–lower abdomen. Further, rectal stimulation showed cortical activation in areas of the brain similar to healthy volunteers. Their assertion is supported by other studies in which SCI patients describe sensation in the bladder,[12,13] genital stimulation[14] and anorectal sensation.[15,16] In another study using MRI, Samdani *et al*.[17] describes a patient who was clinically tested as incomplete, but had complete disruption of their spinal cord. In this patient, sensation below the injury could not have been transmitted via the cord.

In this study, tester variability was reduced by conducting a training session by an expert who has provided training for several multicentered studies.[6] Furthermore, strict criteria were used to determine presence of AS. The manual only specifies a patient 'to describe any sensory awareness, including feelings of touch and/or pressure, when firm pressure with the digit is placed on the rectal walls.' In our study, patients were instructed only to respond positively to sensation of pressure felt rectally, and not to respond to autonomic responses such as goose bumps, tingling, or flushing felt above their injury. To eliminate guessing, each subject had to identify the firm pressure a minimum of three times. The strict criteria and explicit verbal instruction in this study compared with the ISNCSCI manual, most likely reduce the number of false negatives, hence providing a conservative estimate of the examination's negative predictability and sensitivity.

A second possibility is that anal pressure has a lower threshold for perception of sensation compared with LT and PP, and therefore is a more sensitive test for cord completeness. If AS does have a lower testing threshold for incompleteness, our results underlines the importance of testing AS, even if a patient is completely insensate at and below T10.

The lack of literature exploring the reliability and validity of sacral sparing is surprising considering its widespread clinical and research applications. Misclassifications of completeness would have profound effects of current and future research. For example, the inclusion criteria for the recent trial of macrophage injections in acute spinal cord injures required the absence of sacral sparing immediately after their injury.[18] Conversely, the Phase II HP184 drug study was only targeting incomplete injures.[19] If AS alone is not a valid indicator of S4-5 cord function, 40% of subjects in our study would potentially have been denied participation in the first study, and incorrectly included in the later study.

Conclusion
Using the current ISNCSCI, it is important that both S4-5 dermatome and AS be tested to determine sacral sparing. The lower correlation and test sensitivity between AS and S4-5 dermatome testing suggests the need for further validation of the sacral sparing component of the ISNCSCI. The high prevalence of sensation between T10 and S3 in patients with AS and S4-5 sensation but not AS alone, implies the former, but not the later, represents patients that are typically thought of as incomplete injures.

Conflict of interest

The authors declare no conflict of interest.

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