Distribution Agreement

In presenting this thesis or dissertation as a partial fulfillment of the requirements for an advanced degree from Emory University, I hereby grant to Emory University and its agents the non-exclusive license to archive, make accessible, and display my thesis or dissertation in whole or in part in all forms of media, now or hereafter known, including display on the world wide web. I understand that I may select some access restrictions as part of the online submission of this thesis or dissertation. I retain all ownership rights to the copyright of the thesis or dissertation. I also retain the right to use in future works (such as articles or books) all or part of this thesis or dissertation.

Signature:

_____ _____

[Spenser Hunt] Date

Assessing the Ability of the Hoffer Scale of Functional Ambulation to Indicate Mobility Scores on the Pediatric Evaluation of Disability Inventory

By

Spenser Hunt

Master of Public Health

Department of Epidemiology

_____

Cecile Janssens, PhD, MSc, MA

Faculty Thesis Advisor

_____

Katherine Ong, PhD, MA

Field Thesis Advisor

_____

Judy Thibadeau, RN, MN, Health Scientist

Field Thesis Advisor

Assessing the Ability of the Hoffer Scale of Functional Ambulation to Indicate Mobility Scores on the Pediatric Evaluation of Disability Inventory

By

Spenser Hunt

Bachelor of Arts

The University of Michigan,

2014

Faculty Thesis Advisor: Cecile Janssens, PhD, MSc, MA

Field Thesis Advisors: Katherine Ong, PhD, MA,

Judy Thibadeau, RN, MN, Health Scientist

An abstract of

A thesis submitted to the Faculty of the
Rollins School of Public Health of Emory University
in partial fulfillment of the requirements for the degree of
Master of Public Health
in Epidemiology

2016

ABSTRACT

Assessing the Ability of the Hoffer Scale of Functional Ambulation to Indicate Mobility Scores on the Pediatric Evaluation of Disability Inventory

By Spenser Hunt

Background. The interest in research in adult spina bifida and the quality of life for people living with spina bifida across the entire lifespan is growing. As the interest in this field grows, research that focuses on enhancing quality of life for people with spina bifida needs to increase as well. The aim of this study was to assess the ability of the Hoffer Scale of Functional Ambulation to identify participants who score 40 or more points on the PEDI mobility normative assessment.

Methods. This study was conducted using a population of 103 children between 3 years old and 6 years old that was obtained from spina bifida multidisciplinary clinics located in Arizona and Utah. The Hoffer Scale of Functional Ambulation's ability to act as a proxy for the PEDI was tested using PEDI FS normative scores and CA normative scores. Agreement was tested between the Hoffer categories and PEDI normative scores; Pearson's correlation coefficient used to assess the relationship between PEDI normative scores. Sensitivity analyses were conducted to assess which combination of Hoffer categories was best able to identify normative scores≥40. ROC curves were constructed using normative scores≥40 as the outcome and the area under the curve was used to assess the discriminative ability of the model.

Results. Of the 103 participants, 18 were excluded based on missing information in the PEDI mobility assessment. 12 patients had FS normative scores≥40 and 22 patients were found to have CA normative scores≥40. Kendall's Tau-b correlation between the Hoffer categories and PEDI FA normative scores was found to be 0.57 and 0.51 for CA normative scores. Pearson's correlation coefficient between PEDI normative scores was $r = 0.67$. The model comparing complete and community ambulators to all others was best able to identify scores≥40. Area under the curve was approximately 0.91 for PEDI FS normative scores and 0.92 for CA normative scores.

Conclusion. The Hoffer Scale of Functional Ambulation can best identify PEDI normative scores≥40 when participant was categorized as a complete or community ambulator. Further research should be conducted with a more representative population and using a variety of assessments.

Assessing the Ability of the Hoffer Scale of Functional Ambulation to Indicate Mobility Scores on the Pediatric Evaluation of Disability Inventory

By

Spenser Hunt

Bachelor of Arts

The University of Michigan,

2014

Faculty Thesis Advisor: Cecile Janssens, PhD, MSc, MA

Field Thesis Advisors: Judy Thibadeau, RN, MN, Health Scientist, Katherine Ong, PhD, MA

A thesis submitted to the Faculty of the

Rollins School of Public Health of Emory University

in partial fulfillment of the requirements for the degree of

Master of Public Health

in Epidemiology

2016

ACKNOWLEDEMENTS

I would like to offer my most sincere thanks to my faculty thesis advisor Dr. Cecile Janssens for her constant support and insightful advice throughout the entirety of this endeavor. To my field advisors, Judy Thibadeau and Dr. Katherine Ong, I thank you for taking the time you took to guide me into this topic and your continued support throughout both the school year and the summer semester. I would also like to express my thanks to the CDC and the participating spina bifida clinics in Arizona and Utah for allowing me to utilize this dataset to complete this study. To my family and to all those mentioned above, my sincerest thanks is not enough to express my gratitude for your support and encouragement.

TABLE OF CONTENTS

1. Background……1
2. Methods……9
3. Results……13
4. Discussion……16
5. References……21
6. Tables……25
7. Figures……26

BACKGROUND

Spina Bifida Overview

The Centers for Disease Control and Prevention (CDC) reports that every year in the United States around 1,500 babies are born with spina bifida. About 3 children are born with this birth defect for every 10,000 live births [1]. While there are no official numbers, there is an estimated population of 166,000 people living in the United States with spina bifida [2].

Spina bifida manifests itself in many forms. The general definition of this birth defect is the incomplete development of the brain, spinal cord, and meninges [2]. The four spina bifida diagnoses are myelomeningocele, meningocele, closed neural tube defects, and occulta. Spina bifida occulta is the most 'minor' of the four categories that present the least amount of and least severe symptoms. Occulta results in one or more malformed vertebrae with people sometimes being born and growing up without ever realizing they were born with this birth defect. There may be minor pain or other neurological symptoms. The second of the spina bifida diagnoses is known as a closed neural tube defect, such as lipomeningocele, that manifests in a variety of ways such as malformed fat, bone, or meninges. Severity ranges from no symptoms in some to incomplete paralysis and other dysfunctions. The third type is spina bifida meningocele, which is seen when spinal fluid and minges protrude through an opening in the back. There are no neural components present in the malformation and a layer of skin may cover the malformation. Finally, the most severe form of spina bifida is myelomeningocele, where parts of the spinal

cord are exposed through the malformation in the spine. This exposure results in partial or complete paralysis and bladder and bowel dysfunction as well [2].

Although the current research on adults living with spina bifida is scarce, there is research to suggest that young children with physical disabilities stand to gain significant improvements in physical ability and mobility throughout the lifespan from placement in early intervention therapy programs [3, 4].

Infants and toddlers explore the world and increase their knowledge by interacting with the objects around them, sitting and observing their environment, and interacting with the world through locomotion and exploration [4, 5]. These early life interactions are important because they significantly improve mobility capabilities and also, by extension, help develop a child's ability to interact with the world, to interact socially, and to increase their own knowledge [4, 6].

For patients with spina bifida, early intervention therapy programs such as physical therapy to improve mobility or occupational therapy to improve functional ability have the potential to improve long-term function by strengthening otherwise unused muscles. Furthermore, as the human body is malleable, early interventions help to reduce the development of certain compensatory movements that detrimentally affect the body and result in permanent malformation or loss of mobility. Many patients with spina bifida slowly lose their mobility as they age due to the development of these compensatory movements and the lack of muscle use [7, 8]. Early interventions targeted at increasing or maintaining mobility could improve the long-term mobility outcome of spina bifida patients who are more mobile at a young age.

Pediatric Evaluation of Disability Inventory

Some patients with spina bifida would benefit more from different mobility interventions than others depending on the severity of their birth defect and the outcome measure that the doctors wish to aim for [9]. One of the main functions of the Pediatric Evaluation of Disability Inventory (PEDI) is to evaluate the presence or absence of functional delays, including delays to mobility [10]. The PEDI is a clinical assessment tool utilized by researchers to assess and examine the functional and performance abilities of children between the ages of 6 months to 7.5 years. The test is administered to both children and their parents. This assessment tool consists of three domains: self-care, mobility, and social function. The PEDI's measurement scales include the functional skills scale (PEDI FS) that measures a child's ability to perform a certain task and the caregiver assistance scale (PEDI CA) that measures the amount of assistance the child needs when performing the same tasks [11]. PEDI is currently used to identify patients who have birth defects that may result in developmental delays, including spina bifida, and who could benefit from certain interventions.

The validity of the PEDI assessment as a means of determining entry into intervention programs and on predicting the usefulness of interventions has been previously demonstrated. Feldman and colleagues demonstrated the concurrent validity, or the similarity between the target measure and another known measure of a similar construct, of the PEDI when they tested how well the PEDI correlated with the Battelle Developmental Inventory Screening Test (BDIST) [12]. The BDIST and PEDI both examine and assess similar adaptive content areas. Feldman's study

revealed a moderately high correlation between the BDIST and the PEDI self-care and mobility scale scores when testing functional skill levels and caregiver assistance [12].

Nichols and colleagues tested concurrent validity using the BDIST as well and obtained similar results [13]. However, Nichols took their tests for validity one step further. They examined the validity of the PEDI using the Peabody Developmental Motor Scales (PDMS) as well. The PDMS is a different test method that utilizes observation of specific test items that are scored using standardized criteria. Nichols and colleagues found a moderate to high correlation between the PEDI and PDMS. They concluded that while the PDMS appeared to be more sensitive when detecting functional delays, the PEDI is a reliable and valid measure of assessing functional performance in children who have disabilities [13]

Tsai and colleagues highlight the importance of the PEDI in developing an individualized rehabilitation program in a population of children with spina bifida [14]. They found that the PEDI accurately detected differences between participants with myelomeningocele and lipomyelomeningocele; the ability of the PEDI to detect these differences is invaluable in examining the functional abilities of children with spina bifida [14].

PEDI scores can be either normative or scaled scores. The PEDI normative measure is a useful measure for comparing the abilities of a child to the rest of the population of interest. In other words, normative scores are used to compare the abilities of an individual child to the average abilities of children who are considered to be normally developing children. Normally or typically developing children refers

to those children without any functional delays. Children who score 50 (standard deviation: 10, range: 40-60) on the normative scale are considered to be normally functioning. The PEDI scaled measure, on the other hand, is useful when looking at an individual's capabilities alone and over the course of several years. Participants who have lower scaled scores are less mobile or functionally capable than participants who possess higher PEDI scaled scores. Without a child's exact age, results using scaled scores cannot be interpreted as normal or abnormal as a child's scaled score will increase with age [15].

For the current study, we are hoping to identify those participants who are achieving normative scores that are one standard deviation below the mean or greater when compared to normally developing children. For that reason, a cutoff normative score of 40 will be used when identifying those who scored one standard deviation below the mean or better on the PEDI mobility domain.

Hoffer Scale of Functional Ambulation

The Hoffer Scale of Functional Ambulation is a very simple classification method that can be easily assigned by either parents or practitioners [18]. This method is used to determine the functional ambulation level of patients with disabilities or impairments with functional ambulation defined as 'the ability to walk, with or without the aid of appropriate assistive devices safely and sufficiently to carry out mobility-related activities of daily living [19]. Unlike the PEDI, all patients with spina bifida can be assigned Hoffer Scale of Functional Ambulation categories based solely on observation of ability to walk and complete basic functional skills [20].

Generally, the Hoffer Scale of Functional Ambulation categories seen in studies are those used by Vogel and colleagues [18]. They categorized ambulation status according to four categories: community ambulator, household ambulator, therapeutic ambulator, and non-ambulator. An individual is categorized as a community ambulator if he or she is capable of walking both inside and outside for most activities and completing any tasks related to mobility with minimal use of mobility aids like a wheelchair. Household ambulators are individuals who walk inside their own homes or while at school but will otherwise utilize a wheelchair for participation in outdoor activities, participation in certain indoor activities, and for all activities located in the larger community. Household ambulators may not be mobile enough to handle some daily activities without the use of mobility aids. Therapeutic ambulators are those individuals who ambulate for therapy purposes only. Non-ambulators are those individuals who do not ambulate or had been ambulatory for less than a year [18]. Therapeutic ambulators were not considered in the current study being conducted. On the other hand, this study considered those who did not use a wheelchair at all to be complete ambulators and those who could not ambulate to complete functional tasks were categorized as non-functional ambulators.

Multidisciplinary Clinics and Treatment

Spina bifida multidisciplinary clinics are clinics that bring together a number of professionals from different specialties such as neurology, neurosurgery, orthopedics, urology, and physical medicine and rehabilitation that have knowledge and experience with spina bifida. These specialists are all located in one place, thus

facilitating a more organized, comprehensive treatment plan as well as making it easier for patients to organize their own healthcare.

Multidisciplinary clinics or other health centers focused on spina bifida are still rare in the United States. Most states only have a single clinic open in the entire state and some states are left without any multidisciplinary clinic offering appropriate care.

With these centers spread out across the country, parents of children with spina bifida oftentimes must go to specialists who do not have a great amount of knowledge or experience dealing with patients with spina bifida.
Because of this, patients have the potential to miss out on key resources and early interventions that could benefit them throughout their life. To compound the problem, many of the clinics open serve a mostly pediatric population, which leaves adults looking for specialized care without a viable option in many cases [16].

Providing both parents and practitioners with the ability to recognize the need for early intervention when tools such as the PEDI assessment are not available or are not feasible is an important step towards ensuring all patients with spina bifida can be placed into appropriate mobility intervention programs. Researchers have examined and begun to identify proxies for the PEDI assessment in other birth defects such as cerebral palsy. McCarthy and colleagues examined the reliability and validity of three separate measures when trying to accurately measure the health and well being of children diagnosed with spastic cerebral palsy [17]. While researchers have begun to study proxies that increase the ability of practitioners and parents to evaluate health and the need for interventions, there is

no current research that provides a valid proxy for the PEDI assessment in a population with spina bifida.

Summary

Establishing a proxy for the PEDI assessment is one way to circumvent the lack of practitioners specializing in spina bifida care and provide parents with the ability to recognize the need for entry into some form of early intervention. Parents and physicians need a simple proxy that can sufficiently determine whether or not a child is capable of functional ambulation. In short, this tool will need to be able to determine how mobile a child is, as evidenced by how much he or she can walk, as well as how mobile he or she is while completing tasks that involve mobility as compared to a typically developing child.

Due to its simplicity and widespread use, the Hoffer Scale of Functional Ambulation could be an ideal way to identify patients with spina bifida who could benefit from entry into early physical or occupational therapy intervention programs. This paper will explore the following key question: 'Can the Hoffer Scale of Functional Ambulation accurately identify patients who score greater than or equal to 40 points on PEDI mobility FS normative test scores and PEDI mobility CA normative test scores?'

METHODS

Study Design

The Spina Bifida Natural History Project was conducted by spina bifida multidisciplinary clinics located in Arizona and Utah and compiled by the CDC, Rare Disorders and Health Outcomes team (CDC, RDHO). Data collected for this study were expansive and included a measure of executive functioning, bowel and bladder management, detailed information on disabilities and surgical procedures, and parental concerns, in addition to the mobility and ambulation assessments examined in this study [21]. The study's dataset is comprised of children aged between 3 years old and 6 years old. Children were recruited based on enrollment in either the Arizona or Utah spina bifida multidisciplinary center and with parental consent.

The dataset contains an expansive list of variables collected via assessment, hospital records, and parental interview. For this study, data were collected via three primary methods: assessments like PEDI and BRIEF, parental interviews, and from hospital and clinic records. The original dataset contained 103 participants. PEDI was used to assess the mobility and general functioning of participants in this study. Parental interviews were conducted in order to identify various parental concerns on their children's development in comparison to other kids their age. These parental interviews were also used to assess the use of wheelchairs or other mobility aids, the use of bowel or bladder management programs, and the presence of other potential health issues or concerns. Finally, using hospital or multidisciplinary clinic records, demographic information such as socioeconomic

status, medical insurance, race, and gender were collected. Important population characteristics also collected using these records included spina bifida type, other disabilities, as well as history of shunt replacement and surgical procedures.

Variables

Level of spinal lesion in this study was collected differently than seen in previously conducted studies. Previously, studies examining spinal level of lesion used the following levels of spinal lesion: thoracic, upper lumbar, mid-lumbar, lower lumbar, and sacral level lesions [3]. For the present study, participants with a lumbar-sacral level of lesion were considered to have a lower lumbar level lesion [22].

For the purposes of this study, variables were limited to those providing demographic information and those relevant to identifying scores on both PEDI mobility functional skills scores and PEDI mobility caregiver assistance scores. Spina bifida type (myelomeningocele, meningocele, and lipomeningocele), level of spinal lesion, and birth weight give an overall picture of the severity of disability in this population.

Both PEDI mobility FS normative scores and PEDI mobility CA normative scores range from 0-100, with a mean of 50 and a standard deviation of 10. As previously mentioned, normative scores are used to compare a child to children who are considered to be typically developing. A mobility score of 50 represents a child who is as mobile as a normally developing child on the normative scale. For this study, the Hoffer Scale of Functional Ambulation used the following categories: complete ambulators, community ambulators, household ambulators, non-

functional ambulators, and non-ambulators. This categorization was used as opposed to the usual four Hoffer Scale of Functional Ambulation categories used in prior studies: community ambulators, household ambulators, therapeutic ambulators, and non-ambulators [18].

In addition to the variables collected for the original dataset, new variables were generated from these original variables for the purposes of this study. Hoffer scale categories were combined in order to conduct sensitivity and specificity analyses. The Hoffer Scale of Function Ambulation categories were dichotomized as follows: complete ambulators compared to all other ambulators, complete and community ambulators compared to all other ambulators, complete, community, household ambulators compared to all other ambulators, and finally non-ambulators compared to all other ambulators.

Statistical Analysis

Data were analyzed and graphs and plots were generated using Statistical Analysis Software (SAS 9.4, Emory University). In order to determine the characteristics of the study population, descriptive statistics were obtained to summarize demographic information used in identifying the population's characteristics.

How are PEDI normative scores distributed across Hoffer Scale of Functional Ambulation categories? Box and whisker plots were generated to illustrate the distribution of both PEDI mobility FS normative scores and PEDI mobility CA normative scores between and within individual Hoffer scale categories. Agreement between the Hoffer scale and PEDI mobility normative scores was found by

calculating Kendall's Tau-b statistic. Kendall's Tau-b is a test statistic that assesses the level of correlation between two variables [23].

Pearson's correlation coefficient was calculated in order to numerically assess the relationship between the PEDI mobility FS normative scores and the CA normative scores.

The ability of the Hoffer scale of functional ambulation to identify PEDI mobility normative scores greater than or equal to 40 on both FS normative scores and CA normative scores was assessed by generating ROC curves with PEDI normative scores greater than or equal to 40 as the outcome. The area under each ROC curve was calculated as a measure of discriminative ability. Sensitivity and specificity analyses were conducted using the combined Hoffer scale dichotomous variables to assess what combination of Hoffer Scale of Functional Ambulation categories is a good indicator for identifying patients who score greater than or equal to 40 points on both PEDI mobility normative scores.

RESULTS

Of the 103 participants, only those who completed both the Pediatric Evaluation of Disability Inventory mobility domain and obtained both a FS normative score and a CA normative score were included in this study (n=85). Of the remaining participants, the Hoffer Scale of Functional Ambulation considered 25 (30%) patients to be complete ambulators, 24 (29%) were considered to be community ambulators, and 16 (19%) were classified as household ambulators. Participating subjects had a mean PEDI mobility FS normative score of 19.46 with a standard deviation of 16.53 and a mean PEDI mobility CA normative score of 25.11 with a standard deviation of 17.16 [Table 1]. These averages indicate that this group of children as a whole is performing significantly below where normally developing children are performing. These averages are well below the current study's arbitrary cutoff point of 40 points. Twelve participants had PEDI FS normative scores greater than or equal to 40 and twenty-two participants had PEDI CA normative scores greater than or equal to 40. A more complete set of demographic characteristics for subjects not excluded from this analysis is presented in Table 1.

Distribution of PEDI FS and PEDI CA normative scores between and within Hoffer Scale of Functional Ambulation categories shows that complete ambulators have a much higher score distribution for both PEDI FS and PEDI CA normative scores when compared to other patients in this study. Conversely, the distribution of PEDI FS and PEDI CA normative scores is much more similar for the other Hoffer Scale of Functional Ambulation categories [Figure 1a, 1b]. While the distribution of PEDI FS normative scores and PEDI CA normative scores appear to be higher for

community ambulators when compared to household, non-functional, and non-ambulators, the distribution is not as completely distinct as it is when comparing complete ambulators to all other categories. Household, non-functional, and non-ambulators have no distinct distribution when compared to the other Hoffer Scale of Functional Ambulation categories for both PEDI FS and PEDI CA normative scores [Figure 1a, 1b].

Between the Hoffer and PEDI FS mobility normative scores, Kendall's Tau-b correlation was found to have a value of 0.57. Kendall's Tau-b correlation had a value of 0.51 between the Hoffer and PEDI CA mobility normative scores. Agreement indicates a moderate relationship between the Hoffer and both PEDI FS mobility normative scores and PEDI CA mobility normative scores, respectively.

The correlation coefficient between PEDI FS normative scores and PEDI CA normative scores was 0.67 ($n=85$, $p<0.0001$). This coefficient indicates there is a statistically significant, moderate correlation between the two PEDI mobility normative scores. A scatterplot was generated in order to better illustrate the relationship between the two variables [Figure 2]. From the scatterplot and Pearson's correlation coefficient, one can partially explain the similarities between the two box and whisker plots noted previously.

In order to determine the discriminative ability of the Hoffer scale when identifying PEDI mobility normative scores, ROC curves were generated for the Hoffer scale using PEDI mobility normative scores greater than or equal to 40 as the outcome for both PEDI FS and PEDI CA mobility normative scores. Area under the curve was calculated and found to be 0.909 using PEDI FS mobility normative scores

greater than or equal to 40 as the outcome and area under the curve was found to be 0.922 when identifying patients with PEDI CA mobility normative scores greater than or equal to 40 [Figure 3a, 3b].

Finally, sensitivity and specificity analysis was conducted in order to determine which combination of Hoffer Scale of Functional Ambulation categories best indicates participants with scores greater than or equal to 40 for both the PEDI FS and PEDI CA normative scores [Figure 3a, 3b]. The best combination of categories when examining PEDI FS normative scores was determined to be complete and community ambulators compared to all other Hoffer Scale of Functional Ambulation categories. This combination of categories was found to accurately identify all patients with PEDI FS mobility normative scores greater than or equal to 40 and accurately identify patients 49% of the time with PEDI FS mobility normative scores less than 40 (sensitivity: 100%, specificity: 49%). Complete and community ambulators compared to the other Hoffer Scale of Functional Ambulation categories was found to accurately identify all patients with PEDI CA mobility normative scores greater than or equal to 40. 56% of patients with PEDI CA mobility normative scores less than 40 were accurately identified (sensitivity: 100%, specificity: 56%) [Figure 3a, 3b].

DISCUSSION

This study aimed to assess the validity of using the Hoffer Scale of Functional Ambulation as a proxy for the PEDI mobility normative scale when evaluating whether or not children with spina bifida can benefit from some form of early physical therapy or occupational therapy intervention program. The PEDI mobility normative score of 40 was arbitrarily chosen as a cutoff point for the purposes of this study based on the population and the normal level of functioning represented by a PEDI mobility normative score of 50 [15, 16]. There has been no previous research examining what a more appropriate cutoff would be.

The distribution of PEDI mobility normative scores seen in the box-and-whisker plots generated in this study serves to indicate the possibility of the Hoffer Scale of Functional Ambulation to correctly identify certain patients with PEDI mobility normative scores greater than or equal to 40. While those with lesser categorizations of mobility (household ambulators and lower) had no distinct distribution, both complete and community ambulators held distinct distributions in the box-and-whisker plots [Figure 1a, 1b].

The scatterplot and Pearson's correlation coefficient between PEDI mobility FS normative scores and mobility CA normative scores shows there is an expected positive correlation between the two variables. This correlation partially explains the high degree of similarity that can be seen when observing the distribution of PEDI mobility normative scores using box-and-whisker plots.

The ROC curves generated for both PEDI mobility FS normative scores and PEDI mobility CA normative scores showed high discrimination, which was one

indicator of the usefulness of the Hoffer Scale of Functional Ambulation in identifying certain patients with PEDI mobility normative scores greater than or equal to 40 [Figure 3a, 3b]. Further examining the sensitivity and specificity analyses, it was determined that a combination of those patients with a Hoffer Scale of Functional Ambulation categorization of complete or community ambulator was the best combination of categories to correctly identify those patients with PEDI mobility normative scores greater than or equal to 40. The other categories in the Hoffer Scale of Functional Ambulation were not able to accurately identify those patients with mobility normative scores greater than or equal to 40 [Figure 3a, 3b]. Therefore, it was found that, for this population, those patients with PEDI mobility normative scores greater than or equal to 40 that the Hoffer Scale of Functional Ambulation classified as complete or community ambulators could be accurately identified.

Asking yes or no whether or not a patient can walk is oversimplified to serve as a proxy for the PEDI assessment. Using tools that measure functional ambulation will yield results that more correctly reflect the measurement goals of the PEDI mobility domain. While the Hoffer Scale of Functional Ambulation is not a good enough proxy for identifying participants categorized as household ambulators or lower in this population, correctly identifying normative scores greater than or equal to 40 for those participants classified as community ambulators in addition to the complete ambulators is a step in the right direction. Undoubtedly there is a need for easier and standardized proxies that can be used to identify patients who may

benefit from early intervention programs. The Hoffer Scale of Functional Ambulation is one scale that has the potential to be an appropriate proxy.

Without the existence of a standardized and easy to use, general proxy, there is a risk that certain people will be receiving an intervention that would not be beneficial to them and conversely others may not enter into an intervention program when they should be participating. This study shows that the Hoffer Scale of Functional Ambulation is a useful tool for identifying patients with PEDI mobility normative scores greater than or equal to 40 when looking at patients with higher levels of functional ambulation.

Further analyses of interest examining the Hoffer Scale of Functional Ambulation should focus on using different cutoff points to determine how well the scale does when identifying normative scores that are greater or less than 50 or greater or less than 60 points on the PEDI mobility scale. These initial analyses indicate that the Hoffer Scale of Functional Ambulation holds promise when examining patients within certain categories, but further analyses would make these findings more clear.

Limitations in this study include the small sample size used and the fact that only patients born in Arizona and Utah participated. Additionally, patients in this study had a disproportionately large number of lower levels of spinal lesion [Table 1]. Since patients with different levels of spinal lesions often experience different degrees of mobility issues, future studies should aim to gather more patients with higher levels of spinal lesion to better assess the validity of using the Hoffer Scale of Functional Ambulation as a proxy.

The interest in observing, documenting, and improving the health of people with spina bifida across the entire lifespan is steadily increasing. While there is currently a lack of research and scientific evidence documenting health across the entire lifespan in this particular population, there is current evidence that there are steps that can be taken in order to improve long-term mobility and health for young children born with physical disabilities [7]. Furthermore, with only a limited number of doctors and clinics specializing in the treatment and continued care of people with spina bifida, the need for a more simple and widespread tool capable of assessing the need for early mobility interventions in patients with spina bifida is high.

In this population, the Hoffer Scale of Functional Ambulation can potentially serve as a means for identifying normative scores on the PEDI mobility assessment that are greater than or equal to 40 points when evaluating certain patients for entry into early mobility intervention programs. Patients who score one standard deviation below the mean or higher on the PEDI mobility normative score more closely resemble normally developing children when compared to those children who score below 40 points and may benefit more from early mobility interventions. Patients who score less than 40 points on the PEDI normative score may be in need of specialized interventions and the use of a proxy may not be ideal. While generalizing the results of this study based on this small population to the entire country cannot be done, this study suggests that the Hoffer Scale of Functional Ambulation could be one method physicians can easily use to more easily identify patients who may benefit from early intervention programs.

Future studies should focus both on examining the ability of the Hoffer Scale of Functional Ambulation to identify different PEDI mobility normative scores and on discovering or developing even more accurate and simple methods of identifying patients for entry into early mobility intervention program. Furthermore, these analyses involving the Hoffer Scale of Functional Ambulation should be retested using the PEDI-CAT instead of the original PEDI. The PEDI-CAT is a more recent, computerized version of the original PEDI assessment that provides an expanded item bank that covers a wider ranges of ages and abilities [23]. The PEDI-CAT has been deemed to be a valid and acceptable replacement for the old paper-and-pencil PEDI assessment [24].

Parents who have children with spina bifida and do not have easy access to a spina bifida multidisciplinary clinic or some other form of specialized care have a higher risk of missing opportunities that would provide immense benefits to their child. Ensuring that there is some form of easy-to-use and widespread proxy for determining entry into early mobility interventions will benefit the entire family across the child's lifespan. This study is the beginning of finding or developing a suitable proxy but much more research needs to be done to either demonstrate the ability of the Hoffer Scale of Functional Ambulation to be a suitable proxy or discover some other tool that is an appropriate proxy for the more detailed and specialized PEDI assessment.

REFERENCES

1. National Center on Birth Defects and Developmental Disabilities, Centers for Disease Control and Prevention. (n.d.). *Spina bifida data and statistics* [Fact sheet]. Retrieved June 28, 2016, from http://www.cdc.gov/ncbddd/spinabifida/data.html
2. National Institute of Neurological Disorders and Stroke. (n.d.). *Spina bifida fact sheet* [Fact Sheet]. Retrieved July 4, 2016, from http://www.ninds.nih.gov/disorders/spina_bifida/detail_spina_bifida.htm
3. Verhoef, M., Barf, H. A., Post, M. W., Asbeck, F. W., Gooskens, R. H., & Prevo, A. J. (2006). Functional independence among young adults with spina bifida, in relation to hydrocephalus and level of lesion. *Developmental Medicine & Child Neurology*, *48*(2), 114-119.
4. Bergman, J. (n.d.). Aging with spina bifida. Retrieved June 28, 2016, from Spina Bifida Association of Western Pennsylvania website: https://www.sbawp.org/the-stages-of-life/aging-with-spina-bifida/
5. Adolph, K. E., Bertenthal, B. I., Boker, S. M., Goldfield, E. C., & Gibson, E. J. (1997). Learning in the development of infant locomotion. *Monographs of the Society for Research in Child Development*, i-162.
6. Needham, Amy, Tracy Barrett, and Karen Peterman. "A pick-me-up for infants' exploratory skills: Early simulated experiences reaching for objects using 'sticky mittens' enhances young infants' object exploration skills." *Infant Behavior and Development* 25.3 (2002): 279-295.

7. Blauw-Hospers, C. H., & Hadders-Algra, M. (2005). A systematic review of the effects of early intervention on motor development. *Developmental medicine & child neurology*, *47*(06), 421-432.

8. Majnemer, Annette. "Benefits of early intervention for children with developmental disabilities." *Seminars in pediatric neurology*. Vol. 5. No. 1. WB Saunders, 1998.

9. Potter, K., Fulk, G. D., Salem, Y., & Sullivan, J. (2011). Outcome measures in neurological physical therapy practice: part I. Making sound decisions.*Journal of Neurologic Physical Therapy*, *35*(2), 57-64.

10. Haley, S. M., Coster, W. J., Kao, Y. C., Dumas, H. M., Fragala-Pinkham, M. A., Kramer, J. M., ... & Moed, R. (2010). Lessons from Use of the Pediatric Evaluation of Disability Inventory (PEDI): Where Do We Go From Here? *Pediatric physical therapy: the official publication of the Section on Pediatrics of the American Physical Therapy Association*, *22*(1), 69.

11. Knox, V., & Usen, Y. (2000). Clinical review of the pediatric evaluation of disability inventory. *British Journal of Occupational Therapy*, *63*(1), 29-32.

12. Feldman, A. B., Haley, S. M., & Coryell, J. (1990). Concurrent and construct validity of the Pediatric Evaluation of Disability Inventory. *Physical Therapy*,*70*(10), 602-610.

13. Nichols, D. S., & Case-Smith, J. (1996). Reliability and validity of the Pediatric Evaluation of Disability Inventory. *Pediatric Physical Therapy*, *8*(1), 15-24.

14. Tsai, P., Yang, T., Chan, R., Huang, P., & Wong, T. (2002). Functional investigation in children with spina bifida–measured by the Pediatric

Evaluation of Disability Inventory (PEDI). *Child's nervous system*, *18*(1-2), 48-53.

15. Haley, S. M. (1992). *Pediatric Evaluation of Disability Inventory (PEDI): Development, standardization and administration manual*. PEDI Resarch Group.

16. Spina Bifida Association. (n.d.). *Clinics*. Retrieved June 28, 2016, from http://spinabifidaassociation.org/clinics/

17. McCarthy, M. L., Silberstein, C. E., Atkins, E. A., Harryman, S. E., Sponseller, P. D., & Hadley-Miller, N. A. (2002). Comparing reliability and validity of pediatric instruments for measuring health and well-being of children with spastic cerebral palsy. *Developmental Medicine & Child Neurology*, *44*(7), 468-476.

18. Vogel LC, Mendoza MM, Schottler JC, Chlan KM, Anderson CJ. Ambulation in Children and Youth With Spinal Cord Injuries. *The Journal of Spinal Cord Medicine*. 2007;30(Suppl 1):S158-S164.

19. Lam T, Noonan VK, Eng JJ, the SCIRE Research Team. A SYSTEMATIC REVIEW OF FUNCTIONAL AMBULATION OUTCOME MEASURES IN SPINAL CORD INJURY. *Spinal cord*. 2008;46(4):246-254. doi:10.1038/sj.sc.3102134.

20. Alter, A. H. (1973). Functional ambulation in patients with myelomeningocele: MM Hoffer, E. Feiwell, R. Perry, J. Perry, and C. Bonnett. J. Bone Joint Surg.(Am): 55 A: 137–148 (January), 1973. *Journal of Pediatric Surgery*, *8*(5), 854.

21. Gioia, G. A., Isquith, P. K., Guy, S. C., & Kenworthy, L. (2000). *Behavior Rating Inventory of Executive Function: BRIEF*. Odessa, FL: Psychological Assessment Resources.

22. Spina Bifida Association. (n.d.). *Level of spina bifida function*. Retrieved June 28, 2016, from http://spinabifidaassociation.org/wp-content/uploads/2015/07/HOW-SB-LESIONS-IMPACT-DAILY-FUNCTION1.pdf

23. Sen, P. K. (1968). Estimates of the regression coefficient based on Kendall's tau. *Journal of the American Statistical Association*, *63*(324), 1379-1389.

24. Dumas, H., Fragala-Pinkham, M., Haley, S., Coster, W., Kramer, J., Kao, Y. C., & Moed, R. (2010). Item bank development for a revised Pediatric Evaluation of Disability Inventory (PEDI). *Physical & occupational therapy in pediatrics*, *30*(3), 168-184.

TABLES

Table 1. Demographic Characteristics

	Overall (n=103)		Overall (n=103)
Sex		**PEDI FS Normative Scores***	
Male	63	Mean (SD)	19.46 (16.53)
Female	40	Missing	18
		Score ≥ 40	12
Race/Ethnicity		**PEDI CA Normative Scores****	
White	70	Mean (SD)	25.11 (17.16)
Hispanic	25	Missing	18
Asian	4	Score ≥ 40	22
Native American	1		
Unavailable	3		
		Hoffer scores+**	
Birth Weight (kg)		Complete	30
Median (IQR)	3.2 (2.3-3.9)	Community	29
Missing	5	Household	17
		Non-functional	10
Level of Lesion		Non-ambulator	9
Sacral	9	Missing	8
Lumbar-sacral	34		
Lumbar	53		
Thoracic	5		
Unavailable	2		
Spina Bifida			
Myelomeningocele	87		
Lipomeningocele	13		
Meningocele	3		

*FS (Functional Skills) subscale measures patient capability
**CA (Caregiver Assistance) subscale measures level of independence
+Measures level of ambulation with complete being the highest level of ambulation and non-ambulators being the lowest level of ambulation

FIGURES

Figure 1a. PEDI FS Normative Score Distribution Across Hoffer Categories

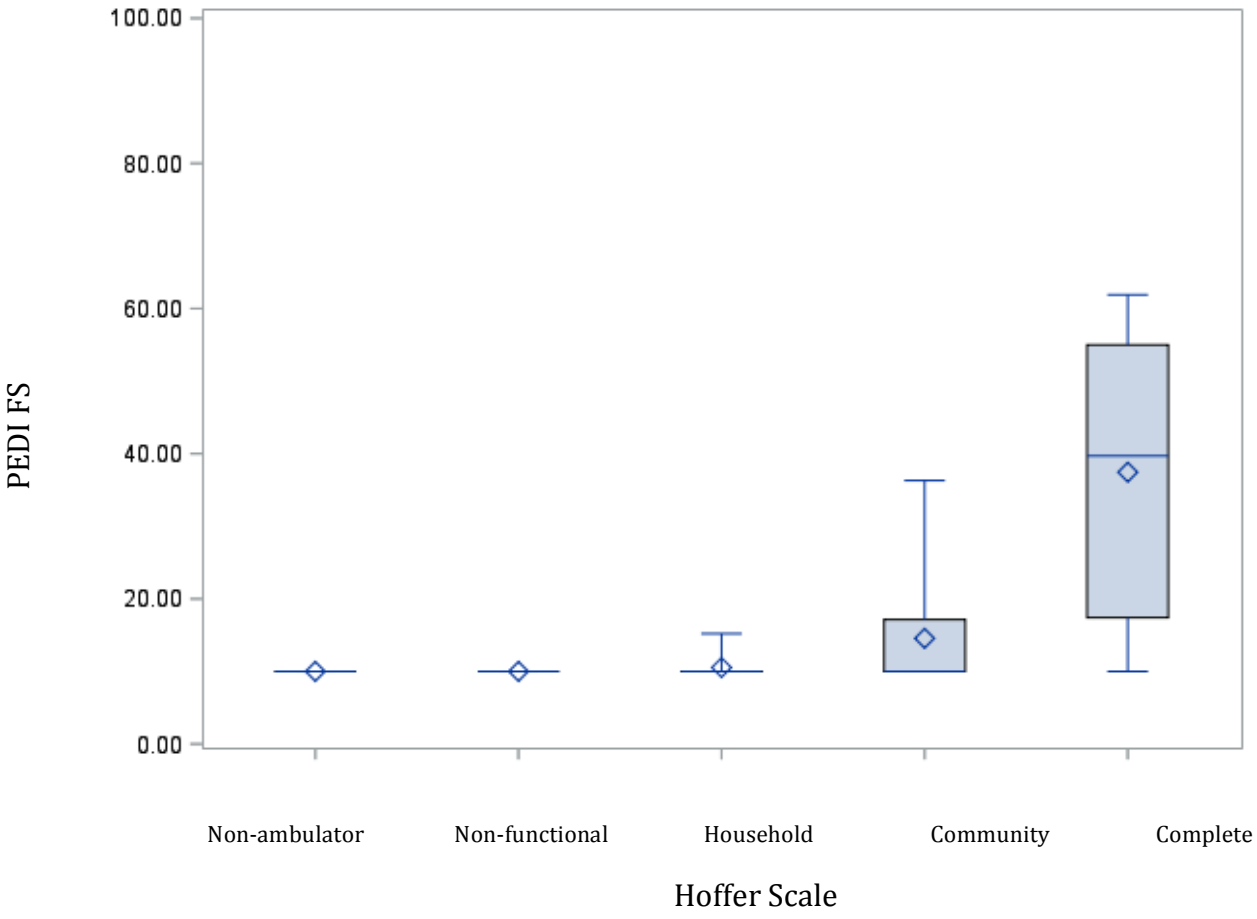

Figure 1b. PEDI CA Normative Score Distribution Across Hoffer Categories

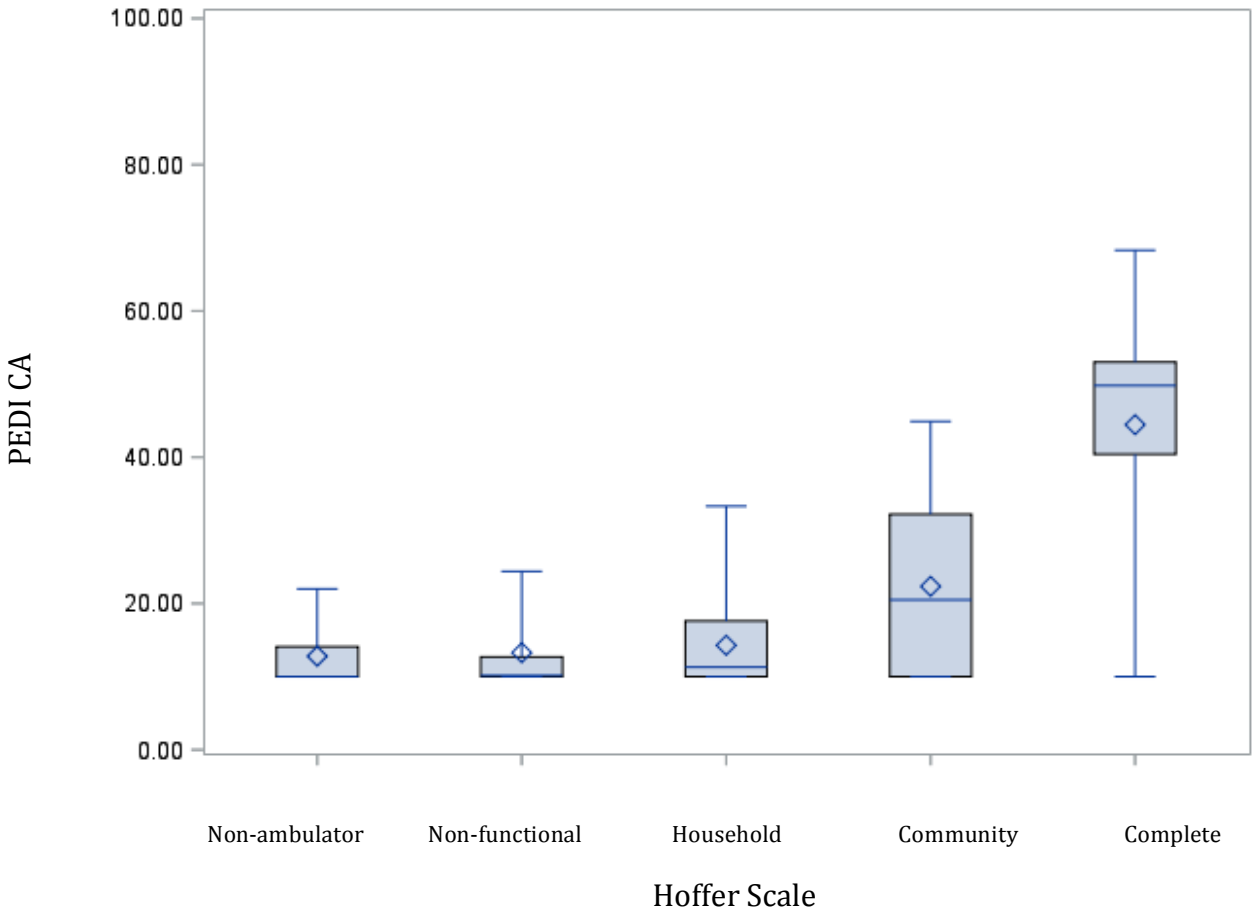

Figure 2. Scatter plot of PEDI Normative Scores in FS and CA Tests

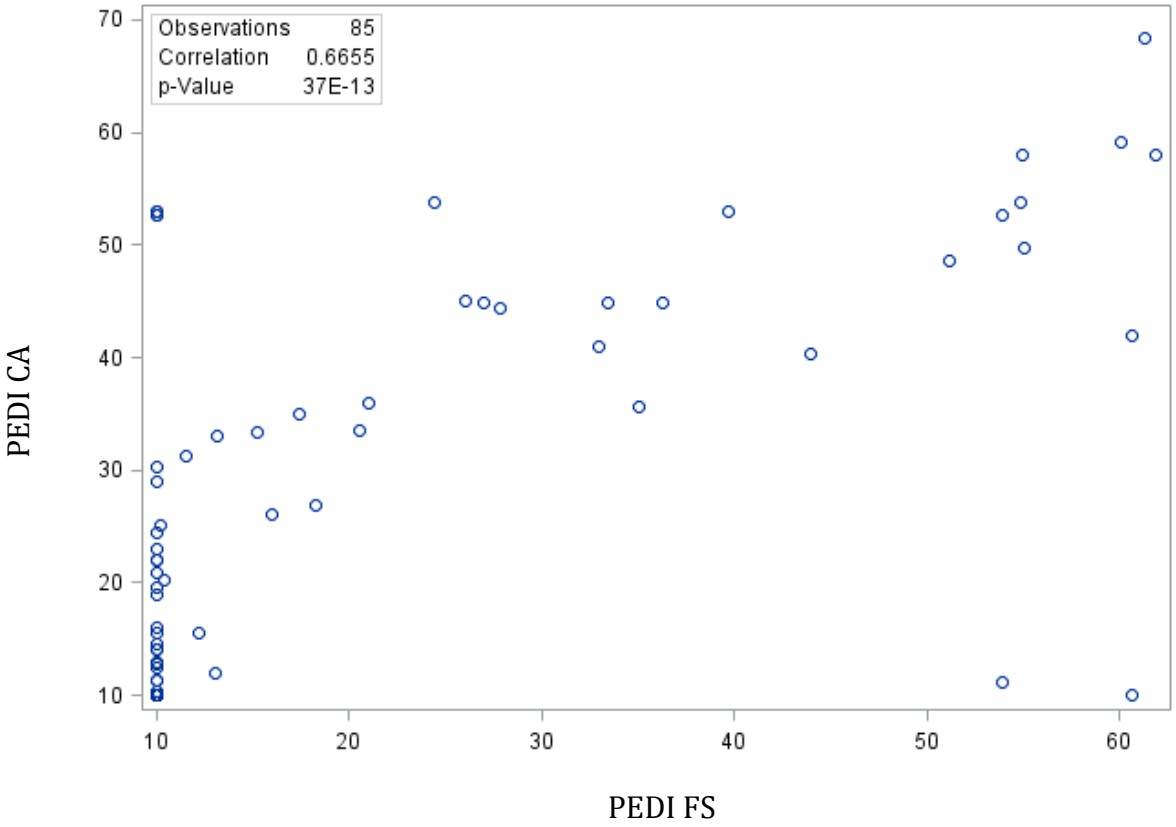

Figure 3a. ROC Curve for Hoffer Scale with PEDI FS Normative Scores≥40 as Outcome

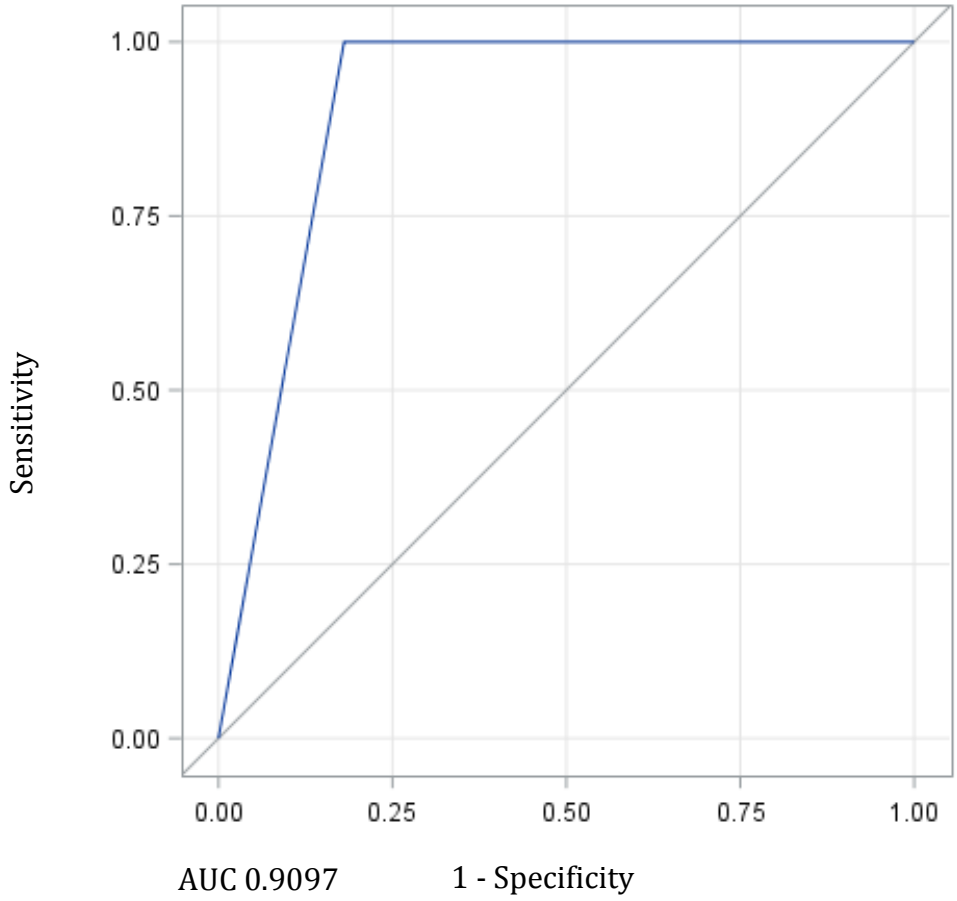

Figure 3b. ROC Curve for Hoffer Scale with PEDI CA normative scores≥40 as Outcome