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Erratum
Musculoskeletal Disorders in Referrals for Suspected Lumbosacral Radiculopathy

ABSTRACT

Objective: An electrodiagnostic evaluation is often requested for patients with suspected lumbosacral radiculopathy. Although musculoskeletal disorders can produce lower-limb symptoms, their prevalence in this referral population is unknown. The purpose of this study was to determine the prevalence of common lower-limb musculoskeletal disorders in patients referred for lower-limb electrodiagnosis and determine whether these findings predict study outcome.

Design: Subjects undergoing an electrodiagnostic study for lower-limb symptoms and suspected lumbosacral radiculopathy constituted the sample. A standardized clinical and electrodiagnostic evaluation was performed for all patients.

Results: There were 170 subjects included. The mean age was 52 (SD = 17), and 45% were males. The total prevalence of musculoskeletal disorders in the sample was 32%. The prevalence in those with a normal study was 55% as compared with 21% in those with lumbosacral radiculopathy (P < 0.0001).

Conclusions: Musculoskeletal disorders are common in patients suspected of having lumbosacral radiculopathy. The high prevalence among both patients with normal studies and those with radiculopathy and other disorders limits the usefulness of this information in predicting study outcome. In particular, it is common for patients to have two or more problems and the presence of a musculoskeletal disorder should not preclude such testing.

Key Words: Radiculopathy, Electrodiagnosis, Musculoskeletal Diseases, Myofascial Pain Syndromes
A large number of conditions are included in the differential diagnosis of patients referred for electrodiagnostic testing. Suspected radiculopathy is a common reason patients are referred to an electrodiagnostic laboratory.\textsuperscript{1,2} Lumbosacral radiculopathy is a pathologic condition involving the lumbosacral nerve roots. The most common symptom is pain that is often increased or precipitated by standing or sitting. Other symptoms that can occur are parasthesias and weakness in a part of the limb. The distribution of the symptoms depends on the nerve roots involved.\textsuperscript{3} Likewise, musculoskeletal conditions are common causes of lower-limb symptoms and may be mistaken for lumbosacral radiculopathy.\textsuperscript{4–7}

Myofascial pain is characterized by pain in skeletal muscles that originates from trigger points, areas within the muscle that are highly sensitive and reproduce the patient’s symptoms when palpated.\textsuperscript{8} Trochanteric bursitis is a condition of inflammation or irritation of either the gluteus maximus bursa or the bursa separating the greater trochanter from the gluteus medius and gluteus minimus. It presents with pain over the lateral aspect of the hip.\textsuperscript{9} Iliotibial band syndrome occurs when the iliotibial band, a dense fascia on the lateral aspect of the knee and hip, impinges over the lateral femoral epicondyle. It most commonly presents with pain over the lateral knee that can radiate up into the lateral thigh or down to the Gerdy tubercle.\textsuperscript{10} In the present analysis, trochanteric bursitis and iliotibial band syndrome are grouped together. Plantar fasciitis is an inflammatory condition of the plantar aponeurosis of the foot. It typically presents with tenderness on palpation of the sole posterior to the calcaneus.\textsuperscript{11}

The purpose of this study was to determine the prevalence of these common musculoskeletal disorders of the lower limb in patients referred for electrodiagnostic testing. In addition, whereas previous studies have examined the influence of physical exam findings on predicting electrodiagnostic study outcomes,\textsuperscript{12,13} in this study the influences of these specific musculoskeletal disorders on electrodiagnostic study outcome prediction were examined.

**METHODS**

**Subjects**

A prospective study was conducted between July 1996 and April 1998 at the following medical centers that treat diverse patient populations and have high-volume Physical Medicine and Rehabilitation electrodiagnostic services: Johns Hopkins University, Baltimore, MD; Ingham Regional Medical Center, associated with Michigan State University, East Lansing, MI; Madigan Army Medical Center, Tacoma, WA; Womack Army Medical Center, Fort Bragg, NC; and Walter Reed Army Medical Center, Washington, DC. Patients were asked to participate if they were referred for lower-limb symptoms or suspected lumbosacral radiculopathy to the electrodiagnostic laboratory in the department of physical medicine and rehabilitation at one of the participating institutions. Subjects participated on a voluntary basis after giving written informed consent. The study protocol was approved by the appropriate institutional review committee at each of the participating institutions.

**Data Collection**

Standardized procedures and data collection sheets were implemented at all participating centers. Patients completed a questionnaire that collected detailed information on symptoms and medical history. For all patients, a standardized physical examination was completed that consisted of the following: (1) a neurologic examination, including manual muscle testing, sensation (vibration and pinprick), and reflex assessments; (2) a musculoskeletal examination; and (3) special tests, such as the straight-leg raise, and the Patrick test. After the questionnaire and physical examination, electrodiagnostic evaluations, conducted by attending physicians and their supervised resident physicians, were performed for all patients. All participating attending physicians were certified by the American Board of Electrodiagnostic Medicine and were skilled musculoskeletal physicians. Such expertise was sought to ensure electrodiagnostic and physical exam findings across participating centers were consistently interpreted.

Musculoskeletal disorders were diagnosed if certain criteria were met in the course of the physical exam. Myofascial pain was diagnosed if palpation of the back or buttock region while avoiding the area of the sciatic notch reproduced symptoms.\textsuperscript{8} Trochanteric bursitis/iliotibial band syndrome was diagnosed if palpation in the lateral hip region reproduced symptoms.\textsuperscript{14,15} Plantar fasciitis was diagnosed if palpation of the sole at the origin of the plantar fascia at the calcaneus reproduced symptoms.\textsuperscript{16}

The standardized electrodiagnostic study consisted of at least (1) one lower-limb motor nerve conduction study, (2) one lower-limb sensory nerve conduction study, and (3) needle EMG with either monopolar or concentric needles of a standard set of 11 muscles. The electrodiagnostic conclusions were classified into three mutually exclusive categories: normal study, lumbosacral radiculopathy, or other condition. A stratified data-collection strategy was used to recruit sufficient persons in each category because these proportions in the referral population were different.
Lumbosacral radiculopathy was diagnosed using methods established earlier. Specifically, lumbosacral radiculopathy was diagnosed if EMG findings (spontaneous activity, increased polyphasias, complex repetitive discharges, or reduced recruitment) were found in two or more muscles innervated by the same nerve root but different peripheral nerves, or if the paraspinal muscles showed spontaneous activity. Overall, 96% of this group had spontaneous activity in at least one muscle. Other diagnoses included sciatic neuropathy, femoral neuropathy, tibial neuropathy, peroneal neuropathy, polyneuropathy, myopathy, and Myasthenia Gravis. The electrodiagnostic outcomes were then analyzed as they related to the presence of musculoskeletal disorders.

Data Analysis

SAS Windows version 9.1 was used to compare the prevalence of musculoskeletal disorders between study groups using χ² analysis or forward two-sided exact test. To further examine the influence of musculoskeletal conditions on study outcome, a multivariate analysis was undertaken using STATA 9.0. Age, gender, and duration of symptoms were controlled for to prevent any potential confounding effects these variables may have had on the outcome of this analysis. Study outcomes were defined as normal study (reference category), lumbosacral radiculopathy, or other diagnosis.

RESULTS

There were 170 subjects included in the study. The mean age of the subjects was 52 (SD = 17) and 45% were men. The mean duration of symptoms reported by the subjects was 26 mos (SD = 54). The prevalence of musculoskeletal disorders is shown in Table 1. Those with a normal study tended to have higher prevalence of the conditions studied. However, persons with electrodiagnostically confirmed conditions also had these disorders, but at lower prevalence than those with normal studies.

The only significant finding from the multivariate analysis showed that the presence of myofascial pain meant about a fivefold lower likelihood of having lumbosacral radiculopathy compared with in a normal study (P = 0.001), given that other variables in the model were held constant.

DISCUSSION

Musculoskeletal disorders are common in patients suspected of having lumbosacral radiculopathy. However, within the study sample there were significant differences in the prevalence of musculoskeletal disorders when subjects were grouped according to the outcome of their electrodiagnostic studies. More than half (55%) of those demonstrating a normal electrodiagnostic study had a musculoskeletal disorder diagnosed on the basis of physical exam findings. In comparison, there was a 21% prevalence of musculoskeletal disorders in those with lumbosacral radiculopathies (P < 0.0001) and a 29% prevalence in those with other electrodiagnostic diagnoses.

Whereas the presence of a musculoskeletal disorder suggests that a patient will have a normal study, the fairly high prevalence of musculoskeletal disorders in those with a confirmed lumbosacral radiculopathy or other electrodiagnostic conclusions limits the usefulness of this information in predicting the results of an electrodiagnostic study. A study should not be curtailed solely on the basis of finding a musculoskeletal disorder on physical exam.

Myofascial pain in particular was common in subjects that had a normal study (39%), significantly more so than in those with a lumbosacral radiculopathy (12%, P < 0.0001) and those with another electrodiagnostic diagnosis (12%, P = 0.0362). This result could be an indication of the

<table>
<thead>
<tr>
<th>Musculoskeletal Disorder</th>
<th>Normal Study, % (n = 51)</th>
<th>Lumbosacral Radiculopathy, % (n = 102)</th>
<th>Other EDX Diagnosis, % (n = 17)</th>
<th>Total Sample, % (n = 170)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Myofascial pain syndrome</td>
<td>39</td>
<td>12*</td>
<td>12*</td>
<td>20</td>
</tr>
<tr>
<td>Trochanteric bursitis/iliotibial band syndrome</td>
<td>29</td>
<td>12*</td>
<td>18</td>
<td>18</td>
</tr>
<tr>
<td>Plantar fasciitis</td>
<td>4</td>
<td>3</td>
<td>18†</td>
<td>5</td>
</tr>
<tr>
<td>One or more of the above</td>
<td>55</td>
<td>21*</td>
<td>29</td>
<td>32</td>
</tr>
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</table>

* Significant at P < 0.05 compared with normal study.
† Significant at P < 0.05 compared with lumbosacral radiculopathy.
‡ Other diagnoses included sciatic neuropathy, femoral neuropathy, tibial neuropathy, peroneal neuropathy, polyneuropathy, myopathy, and Myasthenia Gravis.
difficulty in distinguishing between myofascial pain and radiculopathy and the degree to which the two disorders mimic each other. The referred pain patterns seen in myofascial pain can be very similar to the dermatomal patterns of the spinal nerve roots and are poorly recognized by most practitioners. Therefore, it is possible that many patients being referred for electrodiagnostic evaluations actually have myofascial pain, either alone or along with radiculopathy. If this is the case, an electrodiagnostic evaluation can help clarify the nature of the problem.

Trochanteric bursitis/iliotibial band syndrome showed a significantly lower prevalence in subjects with lumbosacral radiculopathies (12%, P = 0.007) than in those with normal studies (29%). It is possible that this finding reflects difficulty in distinguishing between these conditions and lumbosacral radiculopathy based solely on physical exam findings, as was proposed with myofascial pain above. Both cases emphasize the importance of electrodiagnostic testing in helping distinguish musculoskeletal disorders from lumbosacral radiculopathy when evidence is present for both and treatment strategies for the musculoskeletal conditions are not effective. However, electrodiagnostic studies are not advocated for all patients with musculoskeletal disorders. Rather, the clinician should consider the possibility of radiculopathy or other disorders even in the presence of such conditions.

There are some potential limitations to this study. It is possible that there was heightened suspicion for certain musculoskeletal conditions given the nature of the prospective study and the careful and systematic examination for these conditions. In addition, there are interrater differences in physical examination even for skilled practitioners that could influence our findings. For example, the exact finger pressure used to elicit findings during palpation was not standardized. The underlying pain system and peripheral nervous system physiology is quite complex and interrelated. These systems may augment the pain responses and perceptions in persons with radiculopathies such that physical exam maneuvers are perceived as more painful than they otherwise might be in persons with radiculopathy.

Finally, not all radiculopathies that produce symptoms can be detected by an electrodiagnostic evaluation. For example, radiculopathies that involve only sensory nerve roots will not produce abnormal EMG findings. Also, some radiculopathies may have associated myofascial pain as a component of their disease process. Electrodiagnostically proven radiculopathies with substantial axonal loss may have myotomal pain in the affected muscles that may mimic more typical myofascial pain. Each of these factors may have contributed to some of the data being misclassified with respect to musculoskeletal conditions present, electrodiagnostic conclusions, or both.

The multivariate analysis showed that the presence of myofascial pain meant that a patient was about five times less likely to have lumbosacral radiculopathy than a normal study. This provides a plausible indication of what the results of electrodiagnostic testing would be in a patient diagnosed with myofascial pain.

These findings are also in line with what was concluded in other studies regarding the predictive ability of physical exam on electrodiagnostic outcome. Physical exam findings are important and helpful in guiding the course of an electrodiagnostic study, but are not sufficient to predict the outcome of the study. Similarly, the presence of musculoskeletal disorders gives some indication about what the outcome of an electrodiagnostic study will be but does not provide enough information to definitively predict the results of the study.

CONCLUSIONS

Musculoskeletal disorders are common in patients referred for electrodiagnostic assessment. However, the presence of a musculoskeletal problem does not accurately predict who will have a normal electrodiagnostic study or an electrodiagnostically confirmed lumbosacral radiculopathy with sufficient discriminative ability to curtail the electrodiagnostic evaluation. Whereas there is a significantly higher prevalence in those with a normal study compared with those with lumbosacral radiculopathy, the fairly high prevalence in all groups makes it difficult to predict the outcome of electrodiagnostic testing based on the presence of musculoskeletal disorders.

REFERENCES

Understanding Pain After Traumatic Brain Injury
Impact on Community Participation

ABSTRACT

Objective: To examine the prevalence of pain 1 yr after moderate to severe traumatic brain injury (TBI) and identify predictors from the time of injury. Additionally, factors related to pain at 1 yr after injury were examined along with the impact of pain on community participation.

Design: Prospective cohort study of 146 individuals enrolled during acute inpatient rehabilitation for TBI and community follow-up at 1 yr after injury.

Results: Higher reports of depressive symptoms during inpatient rehabilitation and at 1 yr after injury were significantly related to reports of pain at 1 yr when controlling for demographic and injury characteristics. Being female and nonwhite were also factors related to increased reports of pain. Pain and community participation were significantly related until depression was entered into the model. Depression is a significant factor in the relationship between pain and community participation.

Conclusion: Whereas pain was frequently reported 1 yr after injury, injury-related factors were surprisingly unrelated. Further evaluation of the role that depression plays in the relationship between pain and community participation will be important to determine appropriate management of pain and depression and to optimize participation in individuals with TBI.

Key Words: Pain, Traumatic Brain Injury, Rehabilitation
The experience and explication of pain among individuals with disability is developing into a focus of rehabilitation research. Studies have been conducted to understand the prevalence and impact of pain on individuals with a variety of disabling conditions such as spinal cord injury, amputation, and multiple sclerosis. Pain after traumatic brain injury (TBI) has only been partly examined, but has been noted frequently as an area in need of further research. Much of the existing research has primarily focused on posttraumatic headache, and typically for those with mild TBI. In addition, most studies on pain after TBI have enrolled individuals presenting to outpatient brain injury clinics, rather than prospectively enrolling subjects, which limit the ability to generalize results.

Pain has also been documented as a significant concern after more severe injury. For instance, Walker and colleagues observed 109 consecutive Veterans Administration patients with moderate to severe TBI for 1 yr. Of those patients observed, 38% reported headache during their inpatient rehabilitation, and approximately 50% of those who initially reported headache continued to endorse headache at 1 yr after injury.

Whereas headache has been more widely examined, other painful conditions that can develop after brain injury have not been as frequently evaluated. Such conditions include spasticity, peripheral nerve injury, reflex sympathetic dystrophy, contracture, and heterotopic ossification. A case series conducted on patients presenting to a brain injury clinic found that 58% of individuals with mild TBI and 52% of those with moderate to severe TBI reported chronic pain. Headaches were the most common source of pain for both groups (47 and 34%, respectively), followed by neck/shoulder, upper-limb, and lower-limb pain. Bryant and colleagues observed 96 patients with severe TBI for 6 mo after injury and found that 31% reported chronic pain associated with head, neck, back, and limb pain. Other reports of pain vary in the literature but typically exceed 50% of all patients assessed.

The problems associated with the presence of pain are particularly worrisome for individuals with TBI. In noninjured individuals, pain has been found to interfere with cognition and exacerbate problems with sleep and fatigue and is strongly associated with anxiety and depression, as well as other psychological disorders such as posttraumatic stress disorder. Mooney and colleagues found that poor recovery after mild TBI was explained in large part by depression and pain. Depression was also found to be highly correlated with headache pain at 1 yr after injury.

The aim of the current study was to determine the prevalence of pain at 1 yr after injury using a secondary analysis of a prospectively collected sample and to examine possible factors related to overall pain reports in individuals with TBI. Possible predictors of pain at 1 yr were examined, as well as factors associated with pain at 1 yr. In addition, the relationship of pain, depression, and community participation was explored.

**METHODS**

**Study Population**

One hundred forty-six individuals who completed the SF-36 at their 1 yr after injury evaluation were included in the current study from a total of 202 individuals who were eligible and consecutively enrolled in the University of Washington’s (UW) Traumatic Brain Injury Model System (TBIMS). Twenty-three of the 202 subjects were lost to follow-up, and 33 did not complete the SF-36 because of limited testing time. No significant differences between those who completed the SF-36 and those who did not were found on gender, age, and lowest Glasgow coma scale (GCS) score within 24 hrs of injury. However, those that did not complete the SF-36 were more likely to be nonwhite individuals. Individuals were enrolled between 1998 and 2001 during their inpatient rehabilitation stay. The TBIMS is a longitudinal database and research program sponsored by the National Institute on Disability and Rehabilitation Research. Criteria for enrollment include (1) primary diagnosis of TBI on admission to the inpatient rehabilitation unit, (2) age greater than 16 yrs, (3) arrival at the emergency department within 24 hrs of injury, and (4) receipt of both acute care and inpatient rehabilitation within the Model System of care. Individuals were assessed during their inpatient rehabilitation stay (baseline) and at 1 yr after injury. During inpatient rehabilitation, individuals were not consented or evaluated until they had two consecutive scores of 75 or above on the Galveston Orientation and Amnesia Test (GOAT), which places them out of posttraumatic amnesia. In addition, study examiners discontinued testing if there was any question on validity of responses. The majority of 1 yr after injury evaluations were conducted in person. Alternatively, when an individual was unable to come in person due to distance from the center they were conducted over the telephone. The study was approved by the institutional review board at the University of Washington.

**Measures**

**Demographics**

Age at the time of injury, years of education, gender (female = 1, male = 2), and race (white = 1, nonwhite = 0) were included in analyses.
Injury Characteristics

Severity of injury was measured using the lowest GCS within 24 hrs of injury. GCS measures level of consciousness as indicated by eye opening, verbal response, and motor response to stimuli. Scores can range from 3 to 15, with scores of 3–8 indicating severe injury, scores of 9–12 indicating a moderate injury, and scores of 13–15 indicating a mild injury.\(^{21}\) Scores were prorated if an individual had a score for the best motor component but was missing the eye opening and/or verbal response component of the GCS.\(^{22}\) In the current sample, verbal response was missing in 78% of cases because subjects were intubated at the time of assessment. Eye opening was missing in only one case. Cause of injury was categorized into four groups: (1) motor vehicle related, (2) fall, (3) violence, and (4) all other causes, including pedestrian, bicycle, and sports-related injuries. In addition, the number of associated injuries that occurred at the time of injury was included to examine the impact of non–TBI-related injuries on reports of pain. The total number of injuries, including fracture, amputation, peripheral nerve injury, brachial plexus injury, intraabdominal injury, spinal cord injury, and/or traumatic pneumothorax/hemothorax was calculated.

Functional Impairment

The Functional Impairment Measure (FIM) instrument is a widely used measure of functional status after TBI. This measure includes 18 items that assess level of independence or dependence in six domains of functioning (self-care, sphincter control, mobility, locomotion, communication, and social cognition). Ratings range from 1 (completely dependent) to 7 (independent). This measure was completed for each patient at time of discharge and at 1 yr after injury.

Depression Symptom Severity

The depression subscale, one of the nine primary dimensions from the Brief Symptom Inventory, was used to determine the level of distress or depression.\(^{23}\) This measure was administered during inpatient rehabilitation (baseline) and at 1 yr after injury. The depression subscale has high internal reliability (Cronbach $\alpha = 0.85$).\(^{23}\) Raw scores from the subscale were transformed into $T$-scores using nonpatient adult norms.\(^{16}\) Higher scores indicate more depression.

Community Participation

The Community Integration Questionnaire (CIQ),\(^{24}\) a widely used measure for assessing home and social integration as well as community involvement after TBI, was used to assess participation at 1 yr after TBI. The measure includes questions about home and social integration and productive activity level. Scores range from 0 to 29, with higher scores representing more community participation. Test–retest reliability coefficients range from 0.83 to 0.97.\(^{24}\)

Pain

The two-item Bodily Pain scale of the SF-36 Health Survey (SF-36)\(^{25}\) was used to assess pain.\(^{26}\) This subscale contains two items that assess pain intensity (ranging from none to very severe) and pain interference (ranging from not at all to extremely), which are combined into a composite score of pain severity and transformed into a 0–100 (no pain) scale, with lower scores indicating more pain. The Bodily Pain scale has demonstrated good test–retest reliability (0.78) and validity.\(^{26}\) In addition, it has been found to have good internal consistency among individuals with TBI.\(^{27}\) The composite score was calculated, using the scoring criteria outlined in the SF-36 manual. Norms are also given in the manual from a United States population sample (mean, 75.15, SD, 23.69).\(^{25}\)

Statistical Analysis

Pearson correlation coefficients were calculated among independent and dependent variables to assess relationships among variables. Two linear regressions were conducted to evaluate (1) baseline predictors of pain level reported on the Bodily Pain Index of the SF-36 at 1 yr and (2) potential associated variables, measured at the same time as the level of pain. Baseline predictors included demographic variables (age at time of injury, education, race, and sex), injury characteristics (GCS, number of associated injuries, cause of injury, FIM score at time of hospital discharge), and baseline BSI depression score. Dummy coded indices of each of the cause of injury factors (motor vehicle accident, violence, fall, or other cause, which included pedestrian, bicycle, or sports injuries) were used in this regression analysis. Potential associated variables included baseline demographics as well as the FIM scores, BSI depression scores, and CIQ scores that were completed at 1 yr after injury.

When the above analyses were completed, exploratory analyses were conducted to further elucidate the relationship between pain, depression at 1 yr after injury, and community participation. A series of analyses were conducted to evaluate the impact of depression on the relationship between pain and community participation and involved computation of three regression equations: (1) pain predicting community participation ($\tau$), (2) pain ($\tau$) and depression ($\beta$) predicting community participation, and (3) depression predicting pain...
(α). All equations controlled for the demographic variables.28

RESULTS

One hundred forty-six individuals who completed the Bodily Pain subscale of the SF-36 at 1 yr after injury were included in the study. Prevalence of pain as well as demographic and injury characteristics of this sample are shown in Table 1. The majority were white males with an average age of 36. The average amount of education was 12.4 yrs (range 3–20 yr). Only 27% of individuals reported having no pain (a score of 100 on the bodily pain subscale), and 45% reported no interference with activities from pain. The average pain rating on the Bodily Pain Index was 66.7. Approximately 50% of the sample sustained their injury in a motor vehicle accident. Descriptive statistics for FIM scores and BSI depression scores at baseline and 1 yr after injury, and community participation measured with the CIQ, are also displayed in Table 1. FIM scores and BSI depression scores at 1 yr were significantly higher than at baseline, indicating that functional status improved and depression worsened (P ≤ 0.001 and 0.05, respectively).

Pearson correlations are presented in Table 2. Results suggest that higher reports of pain at 1 yr after injury were significantly related to being female, nonwhite, and having lower function as measured by the FIM instrument at 1 yr after injury. Additionally, higher reports of pain were significantly related to higher rates of depressive symptoms both at baseline and 1 yr after injury, and lower community participation as measured by the CIQ. Interestingly, neither severity of TBI nor number of associated injuries was associated with pain at 1 yr after injury. Higher rates of depressive symptoms at baseline and 1 yr after injury were related to being female, having lower function at 1 yr and lower community participation. Baseline depressive symptoms were also related to lower function at baseline. Reports of higher levels of community participation at 1 yr were related to younger age, being white, having more education, having more injuries, and highly related to better level of function measured by the FIM instrument.

Evaluation of cause of injury suggests that individuals who were violently injured had significantly higher levels of pain than those in the other injury category (mean 50.94 compared with 78.32,

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<th>TABLE 1 Sample characteristics</th>
<th>n</th>
<th>Percentage of Sample</th>
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<tbody>
<tr>
<td>Gender</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>111</td>
<td>76.0</td>
</tr>
<tr>
<td>Female</td>
<td>35</td>
<td>24.0</td>
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<tr>
<td>Race</td>
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<tr>
<td>White</td>
<td>114</td>
<td>78.1</td>
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<tr>
<td>Nonwhite</td>
<td>32</td>
<td>21.9</td>
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<tr>
<td>Cause of injury</td>
<td></td>
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<tr>
<td>Motor vehicle related</td>
<td>71</td>
<td>48.6</td>
</tr>
<tr>
<td>Fall</td>
<td>33</td>
<td>22.6</td>
</tr>
<tr>
<td>Violence</td>
<td>17</td>
<td>11.6</td>
</tr>
<tr>
<td>Other</td>
<td>22</td>
<td>15.1</td>
</tr>
<tr>
<td>Missing</td>
<td>3</td>
<td>2.1</td>
</tr>
<tr>
<td>Mean (SD) Range</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age</td>
<td>146</td>
<td>36.1 (14.9) 17–88</td>
</tr>
<tr>
<td>Education</td>
<td>145</td>
<td>12.4 (2.7) 3–20</td>
</tr>
<tr>
<td>Glasgow coma scale</td>
<td>143</td>
<td>9.2 (3.1) 3–15</td>
</tr>
<tr>
<td>Number of associated injuries</td>
<td>146</td>
<td>2.0 (1.7) 0–7</td>
</tr>
<tr>
<td>FIM scores</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Baseline</td>
<td>144</td>
<td>112.5 (14.4) 32–126</td>
</tr>
<tr>
<td>1 yr after injury</td>
<td>140</td>
<td>122.7 (5.5) 86–126</td>
</tr>
<tr>
<td>BSI depression</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Baseline</td>
<td>132</td>
<td>52.9 (9.6) 36–78</td>
</tr>
<tr>
<td>1 yr after injury</td>
<td>136</td>
<td>55.8 (12.2) 36–80</td>
</tr>
<tr>
<td>CIQ functional score</td>
<td>142</td>
<td>17.7 (6.0) 2.5–29</td>
</tr>
<tr>
<td>SF-36 bodily pain</td>
<td>146</td>
<td>66.7 (28.4) 0–100</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>TABLE 2 Correlations among variables</th>
<th>Bodily Pain Index</th>
<th>Baseline Depression</th>
<th>1-yr Postinjury Depression</th>
<th>CIQ</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age</td>
<td>−0.07</td>
<td>0.10</td>
<td>0.15</td>
<td>−0.21*</td>
</tr>
<tr>
<td>Gender</td>
<td>0.19*</td>
<td>−0.21*</td>
<td>−0.36†</td>
<td>−0.08</td>
</tr>
<tr>
<td>Race</td>
<td>0.19*</td>
<td>0.04</td>
<td>0.01</td>
<td>0.17*</td>
</tr>
<tr>
<td>Education</td>
<td>0.11</td>
<td>−0.13</td>
<td>−0.11</td>
<td>0.28†</td>
</tr>
<tr>
<td>Number of injuries</td>
<td>0.07</td>
<td>0.01</td>
<td>−0.07</td>
<td>0.17*</td>
</tr>
<tr>
<td>Glasgow coma scale</td>
<td>−0.07</td>
<td>−0.08</td>
<td>0.00</td>
<td>0.14</td>
</tr>
<tr>
<td>Discharge FIM score</td>
<td>0.10</td>
<td>−0.17*</td>
<td>−0.02</td>
<td>0.36†</td>
</tr>
<tr>
<td>1-yr FIM score</td>
<td>0.23*</td>
<td>−0.30†</td>
<td>−0.33†</td>
<td>0.50†</td>
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<tr>
<td>Baseline depression</td>
<td>−0.30†</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1-yr postinjury depression</td>
<td>−0.54†</td>
<td>0.50†</td>
<td></td>
<td></td>
</tr>
<tr>
<td>CIQ community participation</td>
<td>0.30†</td>
<td>−0.37†</td>
<td>−0.33†</td>
<td></td>
</tr>
</tbody>
</table>

* P ≤ 0.05; † P ≤ 0.001.
respectively). No other significant differences were found.

Predictors and Correlates of Pain at 1 yr After Injury

Table 3 displays the results of the regression model with the outcome of pain at 1 yr after injury and predictors from baseline, including demographic factors, injury characteristics, function at discharge, and depressive symptoms. Overall, these variables accounted for 25% of the variance in the Bodily Pain measure ($P < 0.001$). Higher levels of depression during inpatient rehabilitation, and being female and nonwhite were associated with higher levels of reported pain at 1 yr after injury, whereas other demographics, function, and community participation were not significantly related to reports of pain when the other factors are taken into account.

Relationship Among Pain, Depression, and Community Participation

Given the finding that community participation was related to reported pain on a bivariate level, but not when included in the regression model, further analyses were conducted to determine how pain, depression, and community participation interrelate. The three regression equations were conducted as described in the Statistical Analysis section, and results are shown in Figure 1. After controlling for demographic variables, pain was associated with community participation. However, when depression was added into the model, the relationship between pain and community participation becomes nonsignificant. This set of relationships suggest that depression is an important variable in the relationship between pain and community participation.

| TABLE 3 Summary of regression analyses for variables predicting pain at 1 yr after injury |
|----------------------------------|--------|--------|-----|
| $B$    | SE $b$ | $t$    |
| Age   | -0.09  | 0.20   | -0.45 |
| Education | 1.14  | 1.07   | 1.08  |
| Race  | 15.87  | 5.79   | 2.74** |
| Sex   | 13.22  | 5.85   | 2.26*  |
| Glasgow coma scale | -0.73  | 0.86   | -0.84  |
| Associated injuries | -1.65  | 1.65   | -1.00  |
| Fall  | -1.80  | 6.54   | -0.27  |
| Violence | -9.90  | 8.28   | -1.20  |
| Other Injury | 7.01  | 7.22   | 0.97   |
| Discharge FIM score | 0.32   | 0.23   | 1.37   |
| Depression at baseline | -0.68  | 0.27   | -2.54* |

$R^2 = 0.25$; adjusted $R^2 = 0.18$. * $P < 0.05$; ** $P < 0.01$.

| TABLE 4 Association with pain at 1 yr after injury |
|----------------------------------|--------|-----|
| $B$    | SE $b$ | $t$    |
| Age   | -0.01  | 0.15   | -0.05  |
| Education | -0.52  | 0.87   | -0.59  |
| Race  | 16.70  | 5.38   | 3.10** |
| Sex   | 3.68   | 5.28   | 0.70   |
| 1-yr FIM score | -0.02  | 0.45   | -0.05  |
| Depression at 1 yr  | -1.10  | 0.20   | -5.46** |
| CIQ   | 0.66   | 0.46   | 1.44   |

$R^2 = 0.36$; adjusted $R^2 = 0.32$. ** $P < 0.01$. 

FIGURE 1 Exploration of the relationship between pain, depression, and community participation. Standardized values are reported in the figure above. $\tau$ and $\beta$ indicate values when only pain or depression predicts CIQ including only demographic variables. $\tau'$ and $\beta'$ indicate the value when both pain and depression are included in the model along with demographic variables.
DISCUSSION

This study shows that pain is a significant problem at 1 yr after injury for individuals with TBI, with 74% of participants reporting some level of pain and 55% reporting interference with daily activities from pain. Whereas these findings suggest higher rates of reported pain than previously published in the literature, the average pain rating of 66.7 is similar to that reported by Findler and colleagues, who found average pain ratings on the Bodily Pain Index of 67 for individuals with moderate to severe TBI. The higher rates of reported pain may be attributable to a variety of factors, including the population included, how pain was assessed, and the development of unmeasured complications (e.g., contractures, heterotopic ossification, and spasticity), which may develop in the year after injury.

Initial evaluation of relationships among variables suggested that pain at 1 yr was correlated with being female, nonwhite, lower functional status, and with higher reports of depressive symptomatology at baseline and 1 yr after injury and with more limited community participation at 1 yr after injury. Similarly, in the regression models with baseline characteristics predicting pain at 1 yr, gender (being female), race (being nonwhite), and higher reports of depressive symptomatology were related to higher pain ratings at 1 yr. Injury characteristics including GCS and number of associated injuries were not found to be related in correlational or regression analyses. This finding is consistent with Walker and colleagues, who report that headache pain was unrelated to injury severity.

At 1 yr after injury, race (being nonwhite) and increased depression were the only variables independently related to higher rates of pain. The significant impact of depression on individuals after TBI has been suggested in the literature and has been found to be related to pain after injury. For example, Mooney and colleagues suggest that poor outcomes after mild brain injury were related to both depression and pain. In addition, Chiu and colleagues found that psychological distress, depression, and poor sleep can lead to lower pain thresholds, which may impact reports of pain. The relationship between being nonwhite and higher reports of pain is difficult to interpret in the current sample, given the small sample size and the heterogeneous population included in the nonwhite category. Future examination of the impact of race on pain reports after TBI is warranted.

Another interesting finding in this study was that community participation was associated with pain on a bivariate level but was not significantly associated with pain in the regression analysis. Further exploration of the relationship among pain, depression, and community participation suggests that depression influences the relationship between pain and community participation at 1 yr after injury. In other words, the relationship between pain and community participation becomes nonsignificant when depression is taken into account. This is a complex relationship that will require further exploration to determine potential causal relationships and the direction of that causality. Although direction of causality cannot be explored with the data available in this study, these results do offer some suggestions for future causal hypotheses. One such hypothesis may be that depression acts as a mediator in the relationship whereby pain leads to increased depression which leads to decreased participation. Alternatively, depression may lead to increased pain reports and decreased participation which would suggest that depression is the influential variable which should be the focus of interest. Whereas reports of depression assessed at baseline and 1 yr were significantly associated with pain at 1 yr after injury, both pain and community participation were only assessed at 1 yr making any determination of causal relationships impossible in the current study. Also, it is possible that variables not measured in this study are important to understanding the relationship among depression, pain, and community participation. Other potential variables of interest may include psychological diagnoses, such as posttraumatic stress disorder, which was found to potentially mediate the relationship between pain and depression and pain and community participation in a sample of patients with severe TBI. Further examination of the possible relationship of depression with pain and function is needed to evaluate the impact of other unmeasured variables.

Results of the current study do suggest that depression and pain are significant problems for individuals with TBI. Further evaluation of the relationship may assist clinicians in determining best treatment approaches. If depression is the key variable leading to increased reports of pain and decreased participation, then treatment for depression should lead to improvements in both. However, if depression is a mediator in the relationship between pain and participation then treatment would need to focus on decreasing pain first, then on treating depression to increase participation. Clinicians would benefit from conducting more detailed evaluations of pain, depression, and functional participation when considering treatment approaches for individuals with TBI such as those described by Martelli and colleagues. Consideration of alternative treatment interventions will be needed given the potential sedative effects of pain medications in individuals with TBI and the
reduced ability to participate in cognitive behav-
ioral treatments. In general, there has been a reluctance to treat pain in cognitively impaired individuals. However, lack of treatment can negatively impact not only individuals with TBI, but their families and caregivers as well. Therefore, further study is warranted to identify where to focus treatment and on the best treatment methods for this population.

This study has several limitations that should be considered in interpreting the results. First, assessments were conducted only at baseline and 1 yr; thus, we cannot determine whether pain reports were ongoing since injury or whether they involved complications developing after TBI. Second, pain evaluation was limited to questions asked on the SF-36, which do not include the site of pain (head vs. body, etc.) or how much pain was associated with other injuries (fractures, amputations, etc.) that occurred at the time of injury. However, both the severity and interference of pain were included. More comprehensive assessment over time may provide information that would determine causal relationships and allow for the evaluation of the role of depression after TBI. In addition, we had no information on whether individuals had been receiving ongoing treatment for their pain or depression or how they were coping with their pain more generally. Third, given the types of measures that were chosen, we have no ability to account for any response bias that may exist and could influence the results. Although it is possible that individuals may have exaggerated their levels of pain and depression, they were instructed as to the confidential nature of their responses, including a certificate of confidentiality. Finally, the proportion of nonwhites was lower in the group that completed the SF-36 compared with those that did not complete it (or were lost to follow up). Individuals who are nonwhite have been found to report higher pain although mixed results in the literature suggest that this finding may be due to other factors.

In the current study, it is not possible to assess whether individuals who participated in the study may differ from those who were lost to follow-up or unable to respond because of other factors. Lesser representation of nonwhites in the sample is likely to underestimate pain. This study was not able to examine the effects of cultural influences on pain and pain perception in TBI patients, but this is an important topic for future study.

The current study suggests that pain is a frequently reported problem at 1 yr after injury in individuals with TBI. Thorough evaluation of pain by clinicians assisting individuals with TBI would be beneficial. In addition, further research is needed to understand the relationship among pain, depression, and community participation to determine causal relationships. Such research may inform clinicians on appropriate assessment and treatment recommendations for this population.

REFERENCES


Myopathic Dropped Head Syndrome
An Expanding Clinicopathological Spectrum

ABSTRACT

Objective: A number of neuromuscular conditions may lead to a dropped head syndrome (DHS), with some patients developing a late onset noninflammatory myopathy affecting only, or predominantly, neck extensor muscles (NEM). The cause, pathogenesis, and nosological classification of this condition are unclear. To further investigate this condition, the authors evaluated the clinical, electrodiagnostic and pathologic findings in seven patients with a myopathic DHS.

Design: Analysis of clinical data, electrodiagnostic studies, and muscle biopsies of seven patients, including one set of identical twins, who developed a very late onset myopathy with severe NEM weakness.

Results: Age of onset was 61–79 yrs, with the pair of identical twins developing NEM weakness within 1 yr of each other (ages 63 and 64, respectively). Seven patients developed weakness (six slight weakness and one more severe) in muscles other than NEM. The group was characterized by the electromyography (EMG) showing a “myopathic” pattern in cervical paraspinal muscles (7/7), muscle biopsies with non-specific myopathic changes on histologic stains (7/7), marked abnormalities in NADH dehydrogenase–reacted sections (6/7), desmin-positive sarcoplasmic deposits (1/7), low carnitine levels by biochemical assays (2/7), and mitochondrial changes (3/7).

Conclusions: Myopathic DHS encompasses a wide spectrum of conditions that strongly affect NEM; however, as documented in the monozygotic twins, some patients may suffer from a distinct, genetically determined form of late-onset restricted myopathy leading clinically to DHS.

Key Words: Dropped Head, Myopathy, Neck Extensor Muscles, Bent Spine
A number of neuromuscular conditions, including neurogenic and myopathic entities, as well as defective neuromuscular transmission, may present with isolated or predominately neck extensor weakness.1-10 Motor neuron disease, polymyositis, and myasthenia gravis are three conditions most commonly associated with severe weakness of neck extensors. The term dropped head syndrome (DHS) was introduced by Suarez and Kelly10 in 1992 to designate patients with a late-onset noninflammatory myopathy that affects only, or predominantly, neck extensor muscles (NEM). Several case series of DHS have been reported.4,7,10-13 Subsequently the term isolated neck extensor myopathy (INEM) was proposed by Katz et al.4,5 to emphasize the distinct clinical presentation of these patients.

The cause and pathogenesis of INEM/DHS remain unclear. It has been increasingly recognized by neuromuscular specialists. The nosological classification of INEM/DHS is controversial, and it likely represents a heterogeneous condition. Some authors have included in the DHS spectrum disorders such as parkinsonism,14 which initially may present with neck extension weakness, adding to the nosological confusion.

We performed a retrospective review of seven patients, including a pair of monozygotic twins, who presented with noninflammatory myopathies with predominant NEM weakness. Our findings reveal that in all cases, the weakness and myopathic changes seen on muscle biopsies extended beyond neck extensor muscle group. It seems that myopathic dropped head syndrome is an expanding clinicopathological spectrum.

PATIENTS AND METHODS

Seven patients were reviewed, including a set of monozygotic twins, who were followed in our neuromuscular clinic spanning the years 1993–2002, and who presented with severe NEM weakness. All patients underwent serial neurological examinations, laboratory studies (including creatine kinase and acetylcholine receptor antibody testing), electrodiagnostic testing (including 2-Hz repetitive nerve stimulation and needle EMG) and muscle biopsy.

Open muscle biopsy sites included the trapezius muscle in two patients, deltoid muscle in four patients, and quadriceps muscle in one patient. The muscle specimens were frozen in liquid nitrogen before being cross-sectioned and stained with hematoxylin and eosin, Gomori trichrome, nicotinamide adenine dinucleotide dehydrogenase-tetrazolium reductase (NADH-Tr), adenosine triphosphatase (at pH 4.3, 4.6, and 9.4), acid phosphatase, nonspecific esterase, periodic acid Schiff, oil red O, and myophosphorylase, per standard protocols.15 Where appropriate, succinate dehydrogenase and cytochrome-c oxidase–reacted sections were also prepared. Quantitative carnitine levels in muscle were obtained in three patients (patients 1A, 1B, and 2). Immunolabeling for desmin was obtained in one patient (patient 4). Electron microscopy (EM) was performed in five patients (patients 1A, 1B, 2, 3, 4).

RESULTS

Clinical Presentation

The clinical and laboratory findings of our seven patients are summarized in Table 1. The age of onset was between 58 and 79 yrs old. Patients typically reported insidious onset of NEM weakness for several weeks to months. The time from symptom onset to diagnosis of DHS was 1–7 yrs. The degree of NEM weakness varied from 1/5 to 5–/5 on the Medical Research Council (MRC) scale. All of our patients experienced at least some degree of weakness in other muscle groups besides the NEM; these frequently included the neck flexors, proximal arm muscles, or proximal leg muscles. The creatine kinase (CK) values ranged from normal to mildly elevated (up to twice the upper limit of normal value).

Patients 1A and 1B were monozygotic twins who developed severe NEM weakness (Fig. 1) at ages 64 and 63, respectively. They have been followed in our neurology clinic for 12 yrs with relative stabilization of their NEM weakness. Patient 2 had, in addition to NEM weakness, mild involvement of shoulder and pelvic girdle muscles. Patient 3 initially presented with NEM weakness but, within 2 yrs, developed cogwheel rigidity in her limbs, a right upper-limb resting tremor, shuffling gait, dysarthria, dysphagia, and limitation in her extraocular eye movements. She was later diagnosed with multisystem atrophy. Patient 4 initially carried the diagnosis of limb girdle muscular dystrophy; he was noted to have accompanying severe weakness of his hip flexors in the range of 1/5 to 2/5 on the MRC scale. Patients 5 (Fig. 2) and 6 developed NEM weakness during several weeks, which then stabilized. No patients had any electrodiagnostic evidence of a neuromuscular transmission defect by 2-Hz repetitive nerve stimulation in several muscle groups, and all patients tested normal for thyroid function and acetylcholine receptor antibody studies. There was also absence of ocular involvement (except patient 3, who developed limitations of extraocular movements late in the course of her disease as part of her diagnosis of multisystem atrophy), speech abnormality, or bulbar signs in our patients on serial neurological examinations.
Magnetic resonance imaging (MRI) of the cervical spine was obtained in all the patients. Aside from what was felt to be age-related degenerative joint disease changes and/or mild disk bulges, there was no significant cervical stenosis, cord compression, bony structural abnormalities, or mass lesions.

**Electrodiagnostic Data**

All the patients had nerve conduction study (NCS) and EMG performed on them at least once.

### TABLE 1 Summary of clinical and laboratory findings

<table>
<thead>
<tr>
<th>Patient</th>
<th>Sex</th>
<th>Age at Onset</th>
<th>Age at Time of Diagnosis</th>
<th>Neck Extension Weakness*</th>
<th>Other Muscle Group Weakness</th>
<th>CK (Normal 0–175 U/liter)</th>
<th>Muscle Biopsied</th>
</tr>
</thead>
<tbody>
<tr>
<td>1A</td>
<td>F</td>
<td>64</td>
<td>65</td>
<td>4 to 5-/5</td>
<td>4+ to 5-/5 neck flexion</td>
<td>352</td>
<td>Trapezius</td>
</tr>
<tr>
<td>1B</td>
<td>F</td>
<td>63</td>
<td>65</td>
<td>4 to 4+/5</td>
<td>5-/5 neck flexion, 4+ to 5-/5 hip flexion</td>
<td>181</td>
<td>Trapezius</td>
</tr>
<tr>
<td>2</td>
<td>F</td>
<td>79</td>
<td>84</td>
<td>2 to 3/5</td>
<td>4/5 shoulder girdle, 4+/5 neck flexion and pelvic girdle and arm abduction</td>
<td>118</td>
<td>Deltoid</td>
</tr>
<tr>
<td>3</td>
<td>F</td>
<td>61</td>
<td>62</td>
<td>1/5</td>
<td>4+/5 neck flexion and arm abduction</td>
<td>369</td>
<td>Deltoid</td>
</tr>
<tr>
<td>4</td>
<td>M</td>
<td>67</td>
<td>74</td>
<td>2/5</td>
<td>4/5 neck flexion and shoulder girdle, 1 to 2/5 hip flexion</td>
<td>44</td>
<td>Quadriceps</td>
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<tr>
<td>5</td>
<td>M</td>
<td>58</td>
<td>59</td>
<td>3/5</td>
<td>4 to 4+/5 neck flexion, 4 to 4+/5 arm abduction, 5-/5 hip flexion</td>
<td>306 (0–215)</td>
<td>Deltoid</td>
</tr>
<tr>
<td>6</td>
<td>F</td>
<td>63</td>
<td>65</td>
<td>3 to 4-/5</td>
<td>4/5 neck flexion, 4+ to 5 neck lateral rotation, 4+/5 arm abduction, 4+ to 5-/5 elbow flexion/extension</td>
<td>569 (0–250)</td>
<td>Deltoid</td>
</tr>
</tbody>
</table>

* Medical Research Council Scale.
CK, creatine kinase.

---

**FIGURE 1** Patients 1A and 1B (identical twins). Both patients were barely able to hold their heads up against gravity (neck extension strength 3/5 on the MRC scale) and eventually required neck collars for support.

**FIGURE 2** Patient 5. Severe neck extension weakness, with dropped head appearance.
Two-hertz repetitive nerve stimulations on the NCS did not reveal any decremental responses to suggest a neuromuscular transmission defect. The EMG showed active denervation (as evidenced by fibrillation changes and positive waves) in five of seven patients (patients 1A, 1B, 2, 4, and 6) and myopathic changes (short duration, small amplitude, and polyphasic motor units) in the cervical paraspinal muscles in seven of seven patients. Of the proximal upper-limb muscles tested, myopathic motor unit potentials were detected in the triceps and deltoid muscles of five of seven patients (patients 1A, 1B, 4, 5, and 6). Four of seven patients (patients 2, 3, 5, and 6) exhibited myopathic changes in the trapezius and infraspinatus muscles. Patient 4, who had severe weakness of his hip flexors (1 to 2/5 on the MRC scale), was found to have myopathic motor units in his iliopsoas and anterior tibialis muscles. Of note, not all patients received uniform screening EMG studies; rather, the examiner directed his study according to the patient’s clinical presentation.

**Muscle Pathology**

The trapezius and deltoid muscles were the most commonly biopsied muscles. One patient (patient 4) underwent a quadriceps muscle biopsy. All muscle biopsies revealed chronic myopathic changes (see Figs. 3 and 4) with large variation in fiber sizes, internalized nuclei, split fibers, and reduced oxidative enzyme activity on NADH-Tr staining. There was no evidence for inflammatory changes. In two patients (patients 1A and 1B), the Gomori trichrome–reacted sections revealed some of the muscle fibers with increased subsarcolemmal accumulations of mitochondria (ragged red fibers), and one of the patients (patient 3) had abnormally shaped mitochondria that contained dense bodies on EM. The muscle biopsies from the monozygotic twins (patients 1A and 1B) revealed some increase in the number of small lipid droplets. Their muscle tissue tested low for total and free carnitine levels (about half of normal for patient 1A and a fifth of normal for patient 1B). The muscle biopsy from patient 4 contained accumulations of sarcoplasmic material that tested positive for desmin (see Fig. 5). Five of the seven muscle biopsies (patients 1A, 2, 4, 5, and 6) revealed mild neurogenic atrophy in addition to the myopathic changes.

**DISCUSSION**

We agree with previously reported postulates that at least a subgroup of patients with a myopathic DHS most likely represent a unique form of restricted late-onset myopathy affecting predominantly NEM. However, our experience with patients diagnosed with myopathic DHS indicates that in some patients, in addition to NEM weakness, there may be coexisting mild involvement of other muscle groups, especially in the shoulder girdle region. This may be easily overlooked clinically when the NEM weakness presents in such a dramatic fashion and the mild degree of weakness in other muscle groups frequently becomes difficult to appreciate. Also, muscle biopsies of the NEM are fraught with technical difficulties as the cervical paraspinal muscle group lies deep under the thick layer of the trapezius. Therefore, alternative muscle biopsy sites usually include the trapezius, deltoid, or biceps. Electrodiagnostic studies with EMG and muscle biopsies of these muscles adjacent to or close to the cervical paraspinals frequently reveal that the myopathic process (not related to inflammatory, metabolic, or advanced dystrophic process) is not purely limited to the...
NEM and represents a more diffuse muscle condition. This was not only observed in our case series but also in some patients from previous reports on DHS.4,10

During the past decade, scattered reports have surfaced linking several disorders (aside from the classic progressive neuromuscular conditions such as polymyositis, motor neuron disease, or myasthenia gravis) with DHS. These include Parkinsonism, chronic inflammatory demyelinating polyneuropathy, inclusion body myositis, and other non-inflammatory myopathies such as nemaline rod myopathy, desmin storage myopathy, mitochondrial myopathy, facioscapulohumeral dystrophy, adult acid maltase deficiency, and carnitine deficiency myopathy.1–5,14,16 Metabolic conditions, such as hypothyroidism and hyperparathyroidism, have also been described in association with DHS.3–5,7,8

Most recently, Rowin et al.17 have reported three patients who received remote (12–36 yrs ago) radiation treatments for Hodgkin’s lymphoma who subsequently presented with DHS. It is imperative that clinicians be vigilant in their evaluations for these conditions as the full clinical picture may be masked for some time. Within our case series, one patient was later diagnosed with multisystem atrophy, two patients (a set of twins) had low muscle carnitine levels, and one patient’s muscle biopsy showed sarcoplasmic accumulations of material that stained positive for desmin. The desmin accumulations may be nonspecific but consistent with a myofibrillar myopathy.18,19 The mitochondrial changes seen in our three patients may only reflect the aging process, rather than a primary mitochondrial myopathy.7,14,20,21

Because all of the patients reported so far have been elderly, one cannot exclude mechanical strain as possibly playing a role in the DHS.4,5,14,21 Many of the patients have documented cervical degenerative joint disease, silent radiculopathies, and kyphosis that may compromise stability, tissue elasticity, and musculoskeletal support of the NEM. These underlying age-related changes may further compound and exacerbate the NEM weakness in genetically susceptible individuals. Although the exact association between these conditions and myopathic DHS has not been well delineated, one should consider that DHS is a more heterogeneous disorder than previously realized.

Work-up for patients with DHS should be tailored according to their clinical presentation, but routine laboratory studies should include CK, erythrocyte-sedimentation rate, C-reactive protein, acetylcholine receptor antibodies, and thyroid studies. Recently introduced to clinical practice, muscle-specific tyrosine kinase antibodies (MuSK Ab) may also be useful in detecting atypical seronegative myasthenia gravis patients presenting with DHS, because electrodagnostic studies may not be sensitive enough to rule out a neuromuscular transmission defect. In a publication by Sanders et al.,22 MuSK Ab were positive in 12 of 32 patients with generalized seronegative myasthenia gravis. Many of these patients exhibited atypical features; for instance, repetitive nerve stimulation was normal in four patients, jitter was normal in six patients, and EMG revealed myopathic motor units in five patients. Three of the 12 patients presented initially with NEM weakness, and if it were not for the presence of MuSK Ab, they very well could have been classified as having DHS consistent with INEM. Although all our patients tested negative for acetylcholine receptor antibody (AchR Ab), MuSK Ab was not yet commercially available to test for any of the patients in our group; thus, atypical myasthenia gravis was not excluded in all of our patients.

Electrodiagnostic testing may detect myopathic change, usually limited to the NEM. Cervical spine MRI should be obtained to evaluate for compressive or spondylitic lesions that may contribute to the DHS. The MRIs of the cervical spine in our patients were not done specifically to evaluate the paraspinous musculature; rather, they were ordered

![FIGURE 5 Patient 4. Foci of abnormal sarcoplasmic material depositions (arrows). A, Modified Gomori trichrome; B, desmin stain. Both ×375.](image-url)
to ensure that there was no significant cervical stenosis, cord compression, bony structural abnormalities, or mass lesions. There have been reports in the literature of edema, atrophy, or fatty replacement of the cervical paraspinal muscles noted on MRI studies. In selected cases, patients may undergo muscle biopsies of clinically or EMG-affected muscles to confirm presence of myopathic changes and to help exclude inflammatory or metabolic conditions. Some authors have performed cervical paraspinal muscle biopsies. However, as already stated, these muscle biopsies may be technically difficult. Therefore, alternative biopsy sites (such as the trapezius, deltoid, or biceps) may be considered, on the basis of clinical and electrodiagnostic findings.

There are no established effective therapies for patients with myopathic INEM. There are anecdotal reports of treatments with steroids, azathioprine, carnitine, coenzyme Q10, and vitamin therapies, but the overall consensus is that these treatments are of dubious utility. The few cases that have reported improvement were patients with inflammatory changes in their muscle biopsies and they were treated with oral steroids, sometimes followed by azathioprine. One can argue that because DHS is felt to be a noninflammatory myopathic process, the inflammation noted on the muscle biopsies of these patients may actually represent focal polymyositis mimicking as DHS/INEM. Patient 5 was placed on a short trial of oral steroids (unknown dose and duration) by his local physician, without apparent improvement.

Six of our seven patients were placed on at least a trial of carnitine supplementation at different doses ranging from 990 mg to 3 g total per day) for at least 6 mos, and although several of them did report improved muscle strength on carnitine supplementation, we could not detect any objective improvement. The monozygotic twins in particular were given carnitine because their muscle carnitine levels were low, but we suspect that the carnitine deficiency may be an epiphenomenon of muscle degeneration rather than representing a primary carnitine deficiency myopathy, because there is evidence that carnitine deficiency does not become a clinically relevant factor in limiting fatty acid oxidation in muscle until carnitine deficiency becomes a clinically relevant factor in limiting fatty acid oxidation in muscle until carnitine deficiency becomes a clinically relevant factor in limiting fatty acid oxidation in muscle until carnitine deficiency becomes clinically relevant the monozygotic twins have not developed significant involvement of other muscle groups, aside from what has been indicated in Table 1. This possibility of a genetic factor in the INEM pathogenesis in some cases may be further supported by some familial cases reported in the bent spine syndrome, a condition with weakness of the truncal extensor muscles that is felt to be within the same spectrum as DHS. Identification of more familial cases and further evaluations with linkage analysis may help elucidate the clinical and genetic spectrum of this unique condition.

REFERENCES


Upper-Limb Prosthetics
Critical Factors in Device Abandonment

ABSTRACT

Objective: To investigate the roles of predisposing characteristics, established need, and enabling resources in upper-limb prosthesis use and abandonment.

Design: A self-administered, anonymous survey was designed to explore these factors. The questionnaire was available online and in paper format and was distributed through healthcare providers, community support groups, and one prosthesis manufacturer. Two hundred forty-two participants of all ages and levels of upper-limb absence completed the survey.

Results: Of participants, 20% had abandoned prosthesis use. Predisposing factors, namely, origin of limb absence, gender, bilateral limb absence, and, most importantly, level of limb absence, proved influential in the decision not to wear prostheses. Enabling resources such as the availability of health care, cost, and quality of training did not weigh heavily on prosthesis rejection, with the exception of the fitting time frame and the involvement of clients in the prosthesis selection. Conversely, the state of available technology was a highly censured factor in abandonment, specifically in the areas of comfort and function. Perceived need emerged as a predominant factor in prosthesis use.

Conclusions: Future research should focus on continued development of more comfortable and functional prostheses, particularly for individuals with high-level or bilateral limb absence. Improved follow-up, repair, and information services, together with active involvement of clients in the selection of prostheses meeting their specific goals and needs, is recommended.

Key Words: Upper Extremity, Limb Prosthesis, Prosthesis Fitting, Prosthesis Design, Rehabilitation
From cosmetic hands, to body-powered hooks, to externally powered devices that flatter their natural counterpart in attempts of mimicry, prostheses design endeavors to address the varying needs and desires of individuals with upper-limb absence. Nevertheless, an estimated 20% of individuals with upper-limb absence reportedly do not use prosthetic devices. Nonuse may be a lifestyle choice based on personal needs, values, and perspectives. It may result from medical conditions that preempt prosthesis fitting. Or, nonuse may be a consequence of external factors such as limited insurance coverage or availability, and inadequate technology. Defining reasons for prosthesis nonuse is pertinent to health professionals, researchers, and administrators involved in the care of individuals with upper-limb absence, the design of improved prosthetics, and the evaluation of clinical strategies.

In a recent review, literature on the motivating factors in prosthesis use and abandonment was explored using the Anderson behavioral model for healthcare use. Under this framework, prosthesis acceptance is modeled as a function of the predisposing characteristics (e.g., gender, level, or origin of limb absence), the established need, and the enabling resources (e.g., healthcare services). The review identifies the following topics as warranting further exploration:

1. The potential roles of specific factors (e.g., origin of limb absence, incidence of pain, fitting time frame) in determining prosthesis use/nonuse.
2. The consequences of prosthesis use/nonuse on overall quality of life and health.
3. The interaction and relative importance of predisposing characteristics, established need, and enabling resources in prosthesis rejection.

Also of importance in forming a realistic picture of prosthesis use and nonuse is the inclusion of nonwearers who, with one notable exception, are often underrepresented because of the tendency to sample only those actively involved with rehabilitation centers. The specific objective of this study was, therefore, to elucidate the motivations for prosthesis use and/or abandonment by a broad examination of the predisposing characteristics, established need, and enabling resources in a population sampled both from hospital- and community-based support networks.

**METHODS**

**Design**

The six-part survey, described in Table 1, was developed in consultation with several individuals with upper-limb absence and a number of prominent researchers and clinicians in the field (see Acknowledgments).

The first four sections were completed by all participants. Section V, exploring reasons for prosthesis abandonment, was completed by nonusers only, and section Vii, regarding prosthesis use and satisfaction, was completed by current or past users who wished to comment on previous prosthetic experiences. The anonymous survey required 20–30 mins to complete and was available online (www.prismlab.org/survey.htm) or in hard copy by contacting the authors.

**Sample Population and Sampling Strategies**

All individuals with upper-limb absence were eligible to participate in this study regardless of (1) level of limb absence, (2) origin of limb absence, (3) bilateral/unilateral limb absence, (4) user status, or (5) types of prostheses used. Parents/guardians were asked to complete the survey for children under the age of 12. The survey was available in English, Spanish, French, and Dutch and was circulated through a number of online support groups (i.e., Arm-Amp, I-CAN, Stumps R Us, UpperEx) and healthcare providers (i.e., Bloorview Kids Rehab, Canada; Shriners Hospital for Children in Los Angeles, CA; Sint Maartenskliniek, the Netherlands). In addition, the survey was promoted on the Otto Bock, Inc., Web site, a prominent manufacturer of upper-limb prostheses. The sample group was self-selected.

**Distribution and Data Collection**

Ten months of data were collected via the online survey. Various measures, as recommended for Web surveys, were implemented to monitor sampling and online data collection:

a. To mitigate nonresponse attributable to spam filtering, incorrect mailing addresses, or mistrust, local collaborators were enlisted to recruit participants. Respondents were asked to specify the organization through which they were recruited.

b. Where possible, prenotification (e.g., mailed flyers) and reminders (e.g., through e-mail or telephone call) were undertaken to increase response rates.

c. A well-established, experienced Web provider (www.vovici.com) was employed to host the survey and to limit any system incompatibilities, such as those introduced by browser settings.

d. The survey was made available in paper format, to reduce sampling bias.
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e. The Internet protocol (IP) address was used to identify and filter repeat respondents.

f. To identify possible false respondents, data were screened for completeness and internal consistency using information patterns (e.g., series of dates, detailed descriptions of limb absence, and prosthesis components).

g. Participant demographics were meticulously collected and reported in an effort to contextualize the sample population and identify possible biases.

Data Analysis

SPSS 15.0 software was used for statistical analyses. Nonuser and user groups were compared via the $\chi^2$ test (with Yates correction where applicable) or the Student’s $t$ test for nominal and interval data, respectively. Differences in ordinal ratings (i.e., satisfaction, design priorities, quality of life, etc.) were evaluated using the Mann–Whitney $U$ test (for two groups) or the Kruskal–Wallis test (for more than two groups). Mean rankings were determined and compared using the Friedman nonparametric ANOVA for repeated measures. Other statistical tools included frequency distributions and three measures of central tendency (i.e., mean, median, mode) as appropriate to the distribution of data in question.

Ethics

Ethical approval for this study was obtained from Bloorview Kids Rehab and the University of Toronto.

RESULTS

Study Population Characteristics

A total of 266 respondents completed the anonymous survey of which 242 were included in this study (i.e., responses were excluded on the following basis: repeated IP address and similarity of data [$n = 3$]; incomplete or inconsistent of data [$n = 7$]; ineligible limb absence [$n = 5$]; under the age of 12 [$n = 9$]). Survey participants were recruited through rehabilitation centers (52%), online support groups (39%), the Otto Bock Web site (3%), or through an independent Internet search or family/friends (6%). Of submissions, 80% were electronic and 20% were by hard copy. The completion rate of the online survey (i.e., the number of surveys submitted divided by the number of times the survey was accessed) was 40%. Previous response rates for surveys addressing the Internet protocol (IP) address was used to identify and filter repeat respondents.

to the wrist, 54% at the transradial level, 21% at the transhumeral level, and 7% at shoulder level or higher. Of participants, 15% had bilateral limb absence. The occurrence of congenital limb absence was higher in the pediatric population (91%) than in the adult population (41%) ($P < 0.001$). Of individuals with acquired limb absence, 54% had lost their dominant hand. Fifty-one percent of participants were male, and 60% were adults (i.e., 19 yrs or older), with an average age of $43 \pm 15$ yrs (19–80 yrs). The average age of the pediatric group ($n = 97$) was 9.5 $\pm$ 6 yrs (1–18 yrs) and, consequently, 70% of pediatric responses were submitted by parents/guardians. Respondents were primarily from Canada (35%), the United States (43%), and Europe (17%).

Of participants, 14% had never worn a prosthesis. Two factors characterized individuals who had never worn a prosthesis: (a) limb absence distal to wrist was more frequent than in the general population ($P = 0.001$), and (b) the mean age of children was younger than in the general population ($P = 0.001$), whereas that of adults was older ($P = 0.002$). According to detailed comments on patterns of use, 28% of participants who had been fitted with a device were categorized as prosthesis rejecters (i.e., used a prosthesis once a year or less), whereas 64% were considered frequent wearers (i.e., either full-time or part-time consistent wear). Of note, 45% of participants recruited from the community rejected prosthetic devices as compared with 21% of those contacted through rehabilitation centers or hospitals ($P = 0.002$). Full-time users wore the prosthesis for an average of $13 \pm 4$ hrs on a typical workday and $10 \pm 5$ hrs on a day off. Part-time users reported prosthesis wear for $7 \pm 4$ hrs per workday and $5 \pm 4$ hrs per day off.

The following will focus on comparisons between prosthesis rejecters and frequent wearers with regards to predisposing factors, established need, and enabling resources.

Predisposing Factors

Level of limb loss

Level of limb absence emerged as a primary predisposing factor in prosthesis acceptance. Specifically, individuals with limb absence proximal to the elbow (high level) or to the wrist (low level) were more likely to reject the device than were those with transradial limb absence ($P = 0.001$).

Origin of limb absence

The relationship between level of limb absence and prosthesis rejection persisted for both congenital and acquired limb absence. Rejection rates for individuals with congenital limb absence were greater for low-level (63%) or high-level (65%)
limb absence and less for transradial (21%) ($P = 0.02$). For acquired limb absence, prostheses were rejected by 0% of individuals with low-level amputations, 16% with transradial, and 39% with high-level amputations ($P < 0.001$). Rejection rates were comparable for individuals with transradial limb absence, regardless of origin. Individuals with acquired amputations at a low or high level seemed less likely to reject prostheses than those with congenital limb absence; however, more data are required for verification.

**Gender**

Overall, females (39%) were more likely to reject a prosthesis than were males (23%), ($P = 0.022$). For males and females with congenital limb absence at any level, no significant differences in prosthesis use were observed. However, females with acquired limb absence were more likely to reject prostheses than were males at both the transradial ($P = 0.042$) and at high levels ($P < 0.001$); not enough data were available to assess low-level limb absence. Females with high-level, acquired limb absence in particular, rejected prostheses in 80% of cases as compared with 15% of males ($P < 0.001$). A high rate of prosthesis acceptance was observed in males with acquired limb absence (91%) as compared with males with congenital limb absence (60%) ($P = 0.001$). The opposite trend was evident in females, for whom 38% with acquired limb absence accepted prostheses, as compared with 70% with congenital limb absence ($P = 0.011$).

**Bilateral limb absence**

In general, rejection rates for bilateral and unilateral limb absence did not differ significantly, irrespective of level of limb absence. Rejection rates for unilateral and bilateral limb absence were comparable in individuals with acquired amputations. However, individuals with congenital, bilateral limb absence had significantly higher rates of rejection (75%) in comparison with those with unilateral limb absence (28%) ($P = 0.004$).

**Length of residua**

The length of residua was not a factor in prosthesis rejection at either the transradial or the transhumeral level.

**Dominant-hand amputation**

Amputation of the dominant hand was not associated with prosthesis acceptance or rejection.

**Medical factors**

Discomfort, ranging from predominantly mild to severe, was reported by participants as follows: residual limb pain (32%), phantom pain (32%), skin irritation (46%), blisters (23%), and upper-body pain (44%). Residual limb pain, phantom pain, and upper-body pain were more prevalent in individuals with acquired limb absence ($P < 0.001$) and in individuals with high-level limb absence ($P < 0.025$). Skin irritation and blisters were more prevalent in frequent wearers than in prosthesis rejecters ($P < 0.001$). No significant differences in residual limb, phantom, or upper-body pain were observed between prosthesis rejecters and frequent wearers, whereas the latter were more likely to experience skin irritation and/or blisters ($P < 0.001$) as consequences of prosthesis use. In general, reports of discomfort and pain were low and the majority did not experience upper-body pain, skin irritation, blisters, residual limb, or phantom pain at any point during a typical week.

**Age**

No differences in prosthesis rejection were observed between adults and children with congenital limb absence, irrespective of gender or level of limb absence. The mean age of prosthesis rejecters and frequent wearers was also not significantly different. However, when grouped by different life stages, as presented in Figure 1, significant differences in rates of rejection were observed ($P = 0.02$), irrespective of origin of limb absence. Rejection rates peaked markedly in three age groups, from 4 to 10 yrs, from 24 to 35 yrs, and for those greater than 65 yrs. These differences are likely related to lifestyle and functional needs, as will be discussed in the subsequent section.

**Established Need**

Participants were asked to rate their perceived need for a prosthesis on a scale from 1 (not at all needed for daily life) to 7 (absolutely essential for daily life). Perceived need varied significantly for frequent wearers and prosthesis rejecters ($P < 0.001$), with median ratings of 6 and 1 for wearers and rejecters, respectively. No difference in perceived need was observed between users of different prosthesis types.

Participants were asked to comment on any activities found to be challenging in everyday life. Of the 178 individuals who responded to this question, 16% had not encountered any insurmountable challenges in their daily life, whereas 10% of individuals found most activities, particularly bilateral tasks, to be challenging. Almost all individuals who reported a great number of challenges made use of a prosthesis to aid in their daily life, whereas frequent wearers and prosthesis rejecters were equally represented in the group not experiencing challenges.

Self-reported activity levels (i.e., use of upper limbs in recreational and job/school activities) and
typical operating environments (i.e., exposure to water, grease, dirt, etc.) were comparable for frequent wearers and prosthesis rejecters. Rates of unemployment and disability leave were low (numbering 3% each) and not statistically different for wearers and rejecters. Figure 2 presents rejection rates for various occupations statuses. Of note, students were more likely to reject prostheses than were full-time workers ($P = 0.021$).

Enabling Factors

Healthcare provision and the state of technology are potential enabling factors in prosthesis acceptance and will be explored in this section.

Health care

Satisfaction with health care in all areas, specifically fitting ($P < 0.001$), follow-up ($P < 0.001$),
repair ($P < 0.001$), training ($P < 0.007$), and information provision ($P = 0.009$), were significantly lower for prosthesis rejecters. Prosthesis rejecters were less satisfied with the information provided with respect to prosthesis technology ($P < 0.001$), sources of funding ($P = 0.01$), use of multiple prostheses ($P = 0.001$), level of expectations set ($P < 0.001$), and the overall knowledge and experience of healthcare providers ($P < 0.001$). Both prosthesis rejecters and frequent wearers were interested in receiving better information on nonprosthetic options (i.e., strategies for accomplishing activities without use of a prosthesis) and resources for peer support. With these two exceptions, frequent wearers were generally satisfied with all other aspects of health care. Prosthesis rejecters reported neutral feelings (i.e., neither satisfied nor dissatisfied) for all other aspects of health care with the exceptions of training and fitting, which elicited median ratings of *satisfied*.

Prosthesis wearers were in much more frequent contact with healthcare providers with regard to upper-limb absence than were rejecters ($P < 0.001$), with appointments occurring at a median frequency of every 7–12 mos, whereas 51% of prosthesis rejecters had not been in contact with healthcare providers for 6 yrs or more.

Of note, satisfaction with overall health care ($P = 0.017$) and the knowledge/experience of healthcare providers ($P = 0.022$) was rated significantly lower by bilateral wearers in comparison with unilateral. Prosthesis wearers with acquired limb absence were less satisfied with all areas of health care and information provision ($P < 0.05$) than were those with congenital limb absence. Individuals with high-level limb absence were less satisfied than those with transradial limb absence with information provided on technology ($P < 0.015$), nonprosthetic options ($P = 0.039$), resources for peer support ($P = 0.008$), and training ($P = 0.027$) and fitting ($P = 0.012$) services.

Satisfaction levels of participants from the three major hospitals involved in this study were comparable in all aspects with the exception of information on funding ($P = 0.003$), which was lower, and information on nonprosthetic options ($P = 0.049$) which was higher, for one hospital. Satisfaction with health care and information provision was significantly lower for individuals recruited from community-based support groups in comparison with the hospitals ($P < 0.005$).

The fitting time frame emerged as an important factor in prosthesis acceptance for individuals with congenital limb absence. Prosthesis rejecters were fitted within a median of 3.9 yrs, with an interquartile range (IQR) of 2–6.6 yrs, whereas frequent wearers were fitted more quickly, within 11 mos (IQR: 5 mos to 1.5 yrs). A similar trend was apparent for individuals with acquired limb absence. Rejecters were fitted at a median 6 mos after amputation, with an IQR of 3 mos to 1 yrs, whereas wearers were fitted within a median of 3 mos (IQR: 2–5 mos). At the time of follow-up, pediatric wearers had worn prostheses for an average of 8 ± 5 yrs, whereas adult wearers had worn them for 21 ± 15 yrs. There was no statistically significant difference between the times of follow-up for prosthesis wearers vs. rejecters.

When asked to rate their involvement in the selection of their primary prosthesis on a scale from 1 (no choice) to 7 (entirely my choice), frequent wearers reported a median rating of 5, whereas rejecters reported a significantly lower degree of involvement of 2 ($P = 0.001$). Prosthesis rejection and involvement in prosthesis selection were not related to the type of device selected, nor was prosthesis rejection.

**Prosthesis technology**

Prosthesis rejecters were significantly less satisfied with all aspects of prosthesis design, including appearance ($P = 0.014$), comfort ($P < 0.001$), function ($P < 0.001$), ease of control ($P < 0.001$), reliability ($P < 0.001$), and cost ($P = 0.034$). The distribution of prosthesis types used by both groups was comparable in terms of the shapes of terminal devices used (i.e., hand or hook) and the mode of actuation (i.e., passive, body powered, or electric). Frequency of maintenance activities (i.e., fitting, replacement of components, and repairs) was also not significantly different between frequent wearers and prosthesis rejecters. Of prosthesis rejecters, 74% stated that they might reconsider prosthesis use if technological improvements were made at a reasonable cost.

**Relative Importance of Factors**

Respondents were asked to specify the importance of a variety of factors in their choice not to wear a prosthesis (a) permanently for the case of nonusers, and (b) on a typical day when the prosthesis is not worn for the case of current users. The percentages of participants who considered each factor to be of some importance in the decision not to wear a prosthesis and the median ratings are presented in Table 2. Evidently, prosthesis rejecters discontinue use largely because of a lack of functional need, discomfort, and impediment to sensory feedback. Discomfort is also a primary factor for occasional nonuse in frequent wearers in conjunction with reasons of necessity (i.e., the prosthesis must be removed for certain activities like swimming or sleeping, for medical reasons, or for repair). Excessive weight was deemed more important by prosthesis rejecters ($P < 0.001$). Heat was considered an important factor in prosthesis non-
wear in both groups, whereas medical factors were considered to be somewhat important by both frequent wearers and prosthesis rejecters in the decision not to wear, as was need for repairs. With the latter three exceptions, prosthesis rejecters rated all other factors of nonwear with greater importance than frequent wearers ($P < 0.001$). Quality of information and training services, along with other enabling factors, such as availability of health care and prostheses, and cost, were generally not considered important in the decision to reject prostheses, although the latter was noted by approximately 50% of individuals as a minor factor in the decision. Technological factors pertaining to prosthesis design were paramount.

**DISCUSSION**

**Summary of Key Findings**

Rejection rates, as determined in this study, were comparable with a meta-analysis of previous literature conducted in this area. Table 3 provides a summary of the key findings with respect to the predisposing factors, enabling resources, and established need. Established need and satisfaction with available prosthesis technology emerged as the predominant factors in prosthesis use and rejection. Individuals who felt that a prosthesis was helpful in their daily activities made use of the device, whereas those who felt it to be a hindrance or unreasonably uncomfortable/difficult to use, did not. It is hypothesized that the decision to wear or reject a prosthesis is resolved in a manner to best meet personal priorities and needs. Although not a primary factor, more than 50% of those surveyed considered availability or cost to play at least a minor role in the decision not to wear a prosthesis.

**Merits and Limitations**

Probably the most interesting attributes of the study presented above are (a) investigation of predisposing factors through exploration of high-order correlations; (b) the ability to assess the relative importance of predisposing factors, enabling factors, and established need on prosthesis use and abandonment; and (c) the opportunity to tap into the opinions and experiences of nonusers recruited outside of the rehabilitation center.

A number of Web-based surveys have been conducted in the past with regards to health care on a large scale (i.e., >100 participants) and

| TABLE 2 Factors in the decision not to wear a prosthesis (a) permanently for prosthesis rejecters or (b) on a specific day for prosthesis wearers, based on self-reported ratings of importance from 0 (not at all a factor) to 3 (most important factor) |
|---------------------------------|-----------------|-----------------|-----------------|
| Factors in Nonwear | Prosthesis Rejecters | Frequent Wearers | |
| Percentage of Respondents | Median Rating | Percentage of Respondents | Median Rating |
| Just as or more functional without it* | 98 | 3 | 60 | 1 |
| More comfortable without it* | 95 | 3 | 66 | 1 |
| Too difficult or tiring to use* | 88 | 2 | 39 | 0 |
| Too heavy* | 83 | 2 | 65 | 1 |
| Too hot | 88 | 2 | 77 | 2 |
| More sensory feedback without it* | 86 | 2 | 40 | 1 |
| Inconvenience* | 93 | 2 | 53 | 1 |
| Lifestyle | 80 | 2 | N/A | N/A |
| Dissatisfaction with prosthetic technology | 70 | 1.5 | N/A | N/A |
| Appearance of the prosthesis* | 72 | 1 | 33 | 0 |
| Medical factors (i.e., skin irritation, blisters, etc.) | 59 | 1 | 64 | 1 |
| Stopped working and needs repair | 49 | 0 | 56 | 1 |
| Cost | 47 | 0 | N/A | N/A |
| Availability of prostheses | 48 | 0 | N/A | N/A |
| Availability of healthcare services | 51 | 1 | N/A | N/A |
| Lack of information about prosthetic options | 27 | 0 | N/A | N/A |
| Lack of training | 28 | 0 | N/A | N/A |
| Someone else made the decision | 15 | 0 | N/A | N/A |
| Moral, cultural, or religious reasons | 8 | 0 | N/A | N/A |
| Must be removed (i.e., for sleeping, swimming) | N/A | N/A | 71 | 2 |
| Mood | N/A | N/A | 51 | 0 |
| Fear of damage | N/A | N/A | 35 | 0 |

* The percentage of respondents who considered the factor to be of some importance in the decision not to wear a prosthesis.

* Factors that were rated significantly more important ($P < 0.001$) by prosthesis rejecters than by frequent wearers.

N/A: As indicated, some factors (e.g. availability, fear of damage, etc.) were not applicable to both prosthesis rejecters and frequent wearers.
prosthetics on a smaller scale (i.e., <100 participants). Internal consistency and test–retest reliability between a large variety of questionnaires administered via the Internet and by other means (i.e., telephone, mail, etc.) have been demonstrated. However, it is important to recognize the possible sampling biases inherent to the results presented herein.

1. The sample population was largely self-selected, as is often the case in consumer-based surveys, making it difficult to assess the extent to which the opinions expressed are reflective of the population as a whole. To quantify this potential limitation, rates of prosthesis rejection in samples recruited from the three primary pediatric hospitals were compared. Response rates for the three hospitals were approximately 40%, 55%, and 100%. No significant differences in rates of prosthesis rejection or satisfaction with overall health care were observed, irrespective of the rate of self-selection.

2. It is expected that the study population based in Canada was, in general, younger, with greater use of electric prostheses than the general population, reflecting the expertise of Bloorview Kids Rehab in myoelectric prostheses and pediatric care. Age (i.e., pediatric or adult) and device type were not factors in prosthesis use or abandonment in either Canada or the United States.

3. To quantify possible sampling biases introduced by the use of an online survey, we compared the demographic distribution of this study’s electronic respondents from the United States (n = 92) with that of a large-scale epidemiologic study (n = 2477), also conducted in the United States in 1996. No statistically significant differences with respect to age, prevalence of transradial limb absence, or origin of limb absence were observed. The prevalence of electric hands and body-powered hooks was not statistically different. In this study, 51% of participants were male, as compared with 63% in the Atkins et al. study (P = 0.02). Previous research has

<table>
<thead>
<tr>
<th>TABLE 3 Summary of key findings</th>
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<tbody>
<tr>
<td>Factor</td>
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<tr>
<td>Predisposing characteristics</td>
</tr>
<tr>
<td>Level of limb absence</td>
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<tr>
<td>Origin of limb absence</td>
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<tr>
<td>Gender</td>
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<td></td>
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<tr>
<td>Bilateral limb absence</td>
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<td>Length of residua</td>
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<td>Dominant-hand amputation</td>
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<tr>
<td>Medical factors</td>
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<tr>
<td>Age</td>
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<tr>
<td>Establish need</td>
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<td>Perceived need for prosthesis</td>
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<td>Occupation status</td>
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<td>Activity levels</td>
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<td>Enabling resources</td>
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found no association between gender and Internet access. It is possible, because of the potential bias of Internet-based surveys to exclude low-income individuals, that concerns regarding high costs of prostheses are actually underestimated in this study. Future research in this area is needed.

The results of this study must be considered in the context of these limitations and should not be generalized to the population en masse.

CONCLUSIONS

Prosthesis use is highly related to established need and enabling resources, particularly the state of available technology. Future research should focus on continued development of more comfortable and functional prostheses, particularly for individuals with high-level or bilateral limb absence. Efforts by healthcare providers to actively involve clients in the selection of a prosthesis most suited to their personal goals and needs should be escalated. Individuals with limb absence are particularly interested in resources for peer support and are desirous to know all of their options, including nonprosthetic. Better support, particularly with regard to information provision, is needed for specialty groups including individuals with bilateral, acquired, or high-level limb absence. Efforts to ensure the availability of quality prostheses and health care should continue.

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A Dynamic Seating Intervention for Wheelchair Seating Discomfort

ABSTRACT

Objective: The objective of this study was to examine the effectiveness of a new user-adjustable wheelchair seating system designed to relieve discomfort for long-duration wheelchair users.

Design: This objective was carried out using the newly developed Tool for Assessing Wheelchair disComfort (TAWC) as the primary outcome measure. Two wheelchair users each tested two different designs and feedback from the wheelchair users regarding the first design was used to guide development of the second design. A single-subject research methodology was used, allowing long-duration (up to 2 wks per test) evaluation of the wheelchair seating systems and comparison of subject discomfort levels with those experienced during a baseline period using their own wheelchairs. The experimental wheelchair seating systems employed existing automotive seating with embedded pneumatic bladders that allowed adjustment of the seat and back-support characteristics. The test wheelchair also had tilt, recline, and elevating leg rests.

Results: The two subjects completed limited periods of testing with the first design, both finding poor results with either stable or increased levels of discomfort. Subject feedback was used to redesign the wheelchair seat. After redesign, both subjects tested the second design and found it substantially more comfortable.

Conclusions: The selected research methodology was a very positive method for a progressive wheelchair seating design and the second design provided improved comfort for both users when compared with that experienced using their own wheelchairs and the first test wheelchair. Future research of this type of user-controlled technology is recommended.

Key Words: Wheelchair Seating, Wheelchair, Discomfort, Disability
Long-duration wheelchair users (i.e., greater than 8 hrs of use per day) with intact sensation frequently experience problematic levels of discomfort. Discomfort has been shown to negatively influence consumer satisfaction, cause decreased quality of life, and lead to problems related to wheelchair propulsion ergonomics and adoption of poor sitting postures—all of which may impair everyday function and the ability to remain seated in a wheelchair. Yet, few researchers have investigated the nature and causes of wheelchair seat discomfort, or possible solutions to this problem. However, the automotive seating industry has done significant product design research to meet the comfort needs of long-duration drivers. Therefore we decided to try and incorporate automotive technologies into wheelchair seating. An additional advantage of this would be the ability to provide lower-cost product options for wheelchair users, because these products are already commercially available in a much larger market than the wheelchair seating market.

Anecdotal experience by clinicians suggests that giving users greater control of the seating system increases satisfaction, but no systematic studies have supported this observation. Preceding work in this research program developed a validated methodology for assessing the affect of wheelchair technology on sitter comfort. The current paper describes the application of this methodology to the development and refinement of an advanced wheelchair seating system. The system, which is based on off-the-shelf commercial components, was intended to be a test bed for technologies rather than a commercial system. As such, the findings reported here may encourage developers to invest in more configurable seating systems for wheelchair users. The purpose of this study was to examine the effectiveness of a this wheelchair seating system designed to relieve discomfort for long-duration wheelchair users.

METHODS

This study involved an iterative process of testing two prototype powered wheelchair seating devices constructed on a Permobil powered wheelchair base. Although the initial seating system design was based on laboratory-based research performed before this study, it was not clear to the researchers that the initial prototype would actually prove to be an effective intervention. The ultimate goal of designing an effective dynamic seating intervention to address the comfort needs of this population of wheelchair users was highly dependent on the outcomes of each design and testing phase. In this study, the initial prototype was tested, but when initial results indicated poor accomplishment of the ultimate comfort-related goal, the study design allowed flexibility to remove the initial prototype from further testing and redesign the seating according to the feedback received from the subjects.

The Permobil powered wheelchair used for testing included powered chair tilt, powered back recline, powered elevating leg rests, and a powered seat lift—all currently available wheelchair seating features. An automotive bucket-style seat was mounted on the Permobil wheelchair base. This seat consisted of a foam contoured seat and back with cloth upholstery. Air bladders were embedded in both the seat and back cushions underneath the foam cushions. Four air bladders were added to the seat, and three bladders were added to the back (see Fig. 1). These air bladders were connected to battery-powered pumps, and the wheelchair user had controls to inflate or deflate each bladder to adjust the support characteristics of the cushion. The wheelchair controls were adjusted until the participant was satisfied and able to independently operate all of the seating and wheelchair controls.

The Tool for Assessing Wheelchair disComFort (TAWC), a previously developed and validated tool, was the main outcome measure, allowing quantification of subject discomfort in a real-life

![FIGURE 1 Arrangement of air bladders in the test wheelchairs.](image)
testing process following a single-system, within subjects research design. Data from the TAWC were used to guide modifications of the wheelchair seating system and to quantify its effectiveness in reducing discomfort. After testing of the first prototype, the seat of the system was redesigned, and the foam structure was replaced with a custom-made, four-quadrant Roho air cushion on the basis of feedback from the participants that the automotive seat was too firm. In addition to the TAWC discomfort scores, subjects monitored their skin daily for any changes in skin integrity for safety. If any redness was detected after sitting in the test wheelchair, the subject was instructed to stop using the wheelchair and contact the researchers. The total amount of time sitting in the wheelchair each day was also monitored via a log completed by the subject each day.

An ABCA single-subject design test protocol was used. Phase A was the baseline phase, during which the subjects used their own wheelchairs. Phase B involved using the prototype wheelchair with only the traditional powered wheelchair features operational. The traditional features used were power seat tilt, power back recline, power elevating leg rests, and power seat lift. Phase C involved using the prototype wheelchair with the traditional wheelchair features as in Phase B, with the new air bladder features operational. Subjects were randomly assigned to either an ABCA or an ACBA test order to minimize possible interference of an order effect.

Two wheelchair users were recruited for this study. The participants reported using powered wheelchairs for an average duration of 8 hrs/day. They had severe motor impairment and intact sensation on their buttocks and lower extremities and experienced discomfort associated with sitting in their wheelchairs. Neither of the participants had experienced skin breakdown on their seating surfaces (buttocks or posterior thighs) within the year before enrollment. Both participants were using Permobil power wheelchairs before the study. The first participant was using a Permobil standing frame power wheelchair when he was initially recruited, but he later received a Permobil power wheelchair with power tilt and recline features. Participant 1 used a high-profile Roho seat cushion, and participant 2 used standard Permobil seating including mildly contoured seat and back cushions.

The TAWC was used to quantify sitting discomfort experienced by the test subjects under all test conditions. This tool is divided into sections that result in two discomfort scores. The General Discomfort Assessment score (GDA) contains eight statements related to discomfort and five statements related to comfort, rated on a seven-point Likert scale. The GDA score has a possible range of 13–91, with higher scores representing increased levels of discomfort. GDA scores were recorded for each 4-hr period of time spent sitting. The Discomfort Intensity Score (DIS) allows subjects to rate level of discomfort in eight body areas and in the body as a whole. The DIS score has a range of 8–99. A score of 8 indicates no discomfort in any part of the body, and a score of 99 indicates a maximum amount of discomfort in eight body areas and in the body as a whole. The reliability and validity of the TAWC have been established and reported in an earlier publication.

Before data collection, participants signed informed consent documents approved by the University of Pittsburgh’s institutional review board. For the baseline measures discussed previously, participants completed daily logs and TAWC questionnaires. The researcher contacted each participant several times per week to answer any questions and clarify procedures as needed. Participants were also encouraged to call or page the researcher if there were any problems or questions regarding either the documentation or the test wheelchair. The researcher visited with each participant weekly to transition the participant from one intervention to the next and to collect data.

Both traditional graphic visual analyses and specialized semistatistical and statistical procedures designed for use in single-subject design were used to analyze the effectiveness of the seating interventions. The comfort effects of three seating systems were compared. The baseline phases (A1 and A2) (participants’ own chairs) were compared with a test chair equipped with traditional powered seating options (B1 and B2) and the same test chair equipped with traditional powered seating options and new user-adjustable seating components (adjustable air bladder systems) (C1 and C2). The two intervention systems were also compared with each other.

All discomfort-related data were manually entered into an SPSS data file, then summary data were transferred into a Microsoft Excel spreadsheet and graphs were developed for performing visual analyses. Semistatistical and statistical procedures used included celeration line analyses and the Tryon C statistic. Before any visual or statistical analysis, the Bartlett test of the lag-1 autocorrelation coefficients was performed on each phase of data collected for each participant. When serial dependency of the data were found, the C statistic alone was relied on for indications of a significant intervention effect, because the C statistic remains effective even with data that are serially dependent.
RESULTS

Both subjects completed partial tests of chair 1. The chair was removed early in both cases because of negative outcomes evidenced by the discomfort scores and development of reddened areas on the buttocks, a preestablished stopping point for safety. Discomfort scores for chair 1 testing are summarized in Table 1. Both subjects also tested chair 2 after the redesign process. This testing allowed comparison of the two chair designs as well as examining the effectiveness of the new design. Table 2 contains chair 2 summary results.

Subject 1

This subject used a Permobil Chairman model powered wheelchair with a high-profile Roho cushion for the A phases of his first trial. He completed phases A, C, and A when testing the first wheelchair design. He had been randomized to an ACBA design, but he experienced redness under one of his ischial tuberosities after 5 days of phase C testing with this first prototype, and therefore the test was ended for safety reasons. He did not progress to phase B; instead, he returned to his own wheelchair for his return to baseline phase (phase A). His GDA and DIS discomfort scores changed little while he used the prototype wheelchair. The scores were slightly lower, but they were not particularly stable, and none of the semistatistical tests indicated a significant difference when using the test wheelchair. The C statistic analysis indicated significantly greater discomfort levels on both the GDA and DIS measures when he was using the first test wheelchair.

After the redesign of the test wheelchair, he was contacted and asked whether he would be interested in testing the redesigned wheelchair. He agreed, but because he had just obtained a new personal wheelchair, he underwent a new trial, including new baseline-phase data collection. His new wheelchair was also a Permobil power wheelchair base, but instead of a standing feature, his new wheelchair had power seating options including power seat tilt and power back recline. He was assigned the same testing schedule: ACBA, for consistency with his first round of testing.

Once again, his mean GDA and DIS discomfort scores changed little across phases. Trend lines illustrated increasing levels of discomfort during all phases of testing. However, the slopes of the trend lines were slightly decreased during phases B and C. On return to baseline, mean discomfort increased and the trend reversed in slope, indicating increasing discomfort when he returned to using his own wheelchair. The DIS data C statistics indicated significantly less discomfort during phase B ($z = 2.55$) and significantly more discomfort during phase C ($z = 2.78$) and his final baseline phase ($z = 2.37$). This indicates lower levels of discomfort with the test wheelchair with traditional power seating, but no further decrease in discomfort with the addition of the new, user-adjustable seat features.

Subject 2

The second subject was randomized to an ABCA testing order. During his baseline phases, he used a Permobil power wheelchair with the standard Permobil foam seat and back cushions. His own wheelchair had power seat tilt, power back recline, and power elevating leg rests. His first chair trial was interrupted during phase B because of complaints of increased discomfort and difficulty using chair 1 in his home environment.

This participant’s mean discomfort scores indicated greater discomfort during phase B with the first chair than during his baseline phase (phase A).

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Chair 1 testing: means and standard deviations of subject discomfort scores</th>
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<tbody>
<tr>
<td>Subject</td>
<td>Phase</td>
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<tr>
<td></td>
<td>A1</td>
</tr>
<tr>
<td>GDA</td>
<td></td>
</tr>
<tr>
<td>score</td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>57.1 (6.4)</td>
</tr>
<tr>
<td>2</td>
<td>39.6 (9.3)</td>
</tr>
<tr>
<td>DIS</td>
<td></td>
</tr>
<tr>
<td>score</td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>33.8 (5.1)</td>
</tr>
<tr>
<td>2</td>
<td>12.7 (1.0)</td>
</tr>
</tbody>
</table>

GDA, General Discomfort Assessment score: 13–91, with higher scores indicating greater discomfort; DIS, Discomfort Intensity Score: 8–99, with higher scores indicating greater discomfort.

<table>
<thead>
<tr>
<th>Table 2</th>
<th>Chair 2 testing: means and standard deviations of subject discomfort scores</th>
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<tr>
<td>Subject</td>
<td>Phase</td>
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<tr>
<td></td>
<td>A1</td>
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<tr>
<td>GDA</td>
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<tr>
<td>score</td>
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<tr>
<td>1</td>
<td>50.2 (3.0)</td>
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<td>2</td>
<td>39.6 (9.3)</td>
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<tr>
<td>DIS</td>
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<td>score</td>
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<tr>
<td>1</td>
<td>21.4 (1.6)</td>
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<td>2</td>
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</table>

GDA, General Discomfort Assessment score: 13–91, with higher scores indicating greater discomfort; DIS, Discomfort Intensity Score: 8–99, with higher scores indicating greater discomfort.
Trends in discomfort levels indicated stable levels of discomfort during both baseline phases and increasing levels of discomfort during phase B. DIS score trends followed a similar pattern as those found with GDA scores with increasing discomfort levels during phase B (see Fig. 2). The result of the $C$ statistic testing of DIS scores for phase B indicated a significantly higher discomfort level ($z = 2.53$) with this intervention.

After redesign of the test wheelchair, this participant agreed to complete his trial using chair 2. Because his own wheelchair had not changed, the test wheelchair was reintroduced, and he completed phases B and C and then returned to using his own wheelchair for his final baseline phase. Celeration line testing of the GDA scores indicated greater discomfort during phase C than during phase B. Overall, GDA scores were quite variable, but with a consistent pattern each day: lower scores in the morning, and higher scores at the end of each day. DIS scores for this participant were slightly less variable overall, yet they still exhibited a similar pattern. Celeration line testing of these scores indicated lower discomfort during phases B and C, but greater discomfort during phase C when compared with B. Figure 2 illustrates the DIS score means and trends. $C$ statistic testing of the DIS scores from phase B with the first wheelchair and phase B with the second wheelchair was significant ($z = 2.69$), indicating a significantly lower discomfort level with chair 2. This was indicative of improved comfort with chair 2 when compared with Chair 1 with the same features.

**DISCUSSION**

The newly developed TAWC subjective assessment tool and associated clinical measures were used in an iterative testing process to develop a prototype wheelchair seating system designed to minimize discomfort of wheelchair users. Chair 1, the first prototype, underwent initial testing by both participants. The TAWC measurements demonstrated that the chair 1 was not providing the expected benefits. Because of the results from the initial testing, the wheelchair seating system was redesigned and then retested as chair 2. The same participants tested chair 2 after this redesign, and the TAWC was again used to assess the effectiveness of the new intervention.

One concern with using a within-subjects design in research is autocorrelated data. Because of this concern, all data were tested for autocorrelation. In these two subjects only data from two phases resulted in autocorrelated data. For these results, the $C$ statistic was relied on for testing of significant results. The remainder of the data were
not autocorrelated, allowing for both visual and semistatistical analysis with all included methods.

The results of this testing must be considered in light of the limitations of this particular research. The difficulties involved in introducing a novel wheelchair design to these participants cannot be overlooked. The wheelchair base itself, which was not being studied, may have caused enough difficulty for the subjects to interfere with the ability to truly examine the effects of the seating intervention. The fit and function of the wheelchair seat also was suboptimal for the participants. Because of the close relationship between fit and comfort, this may have caused suboptimal levels of comfort for the subjects unrelated to the dynamic feature effects.

Recommendations for future study include (1) studying “standalone” seating products intended for use in the participants’ own wheelchairs; (2) reducing the TAWC scoring to twice per day—once at 6 hrs and once at bed time—because of the relatively low levels of discomfort reported with shorter sitting durations; (3) reducing the data-collection labor involved on the part of the participants (perhaps by automating more of the data-collection process) and extending the phase lengths to 8–10 days per phase to attain greater stability of trends; and (4) increasing the number and pattern of alternating phases—for example, studying one intervention at a time and using an ABABA design.

CONCLUSION

The goal of this research was to assess the effectiveness of a new, user-adjustable wheelchair seating system in alleviating discomfort. A secondary goal was to use the subject data in further development of the wheelchair seating design. These goals were accomplished through this within-subjects methodology. The first wheelchair design, developed after short-duration laboratory testing, did not meet the goal of reducing seating discomfort with long-duration sitting. Therefore, this wheelchair was recalled and redesigned. On construction of the new design, the wheelchair was reintroduced and retested by these two participants. This method of testing was very helpful in designing a wheelchair that was able to meet the goal of enhancing comfort of users from the target population. The within-subjects design allowed for in-depth investigation of the effectiveness of a user-adjustable seating system for long-duration wheelchair users with intact sensation, using three measures of discomfort.

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Cyproheptadine for Pediatric Intrathecal Baclofen Withdrawal
A Case Report

ABSTRACT


Intrathecal baclofen withdrawal syndrome is a known complication of intrathecal baclofen pumps. Its origin is postulated as an independent form of a serotonergic syndrome occurring from loss of \( \gamma \)-aminobutyric acid B receptor–mediated inhibition of serotonin. Prodromal symptoms include pruritis, a return of deep tendon reflexes, and increased spastic hypertonia. Previous reports have documented use of cyproheptadine in treatment of this syndrome in adults with positive results. We present the case of a 14-yr-old child with cerebral palsy who developed pruritis and worsening spastic hypertonia 18 mos after pump implantation. She had been previously treated with 520 \( \mu \)g/day of intrathecal baclofen. Progression of her symptoms was successfully arrested by the administration of both oral and intrathecal baclofen and 6 mg of oral cyproheptadine every 6 hrs for 1 day. We postulate that cyproheptadine should be considered a valuable adjuvant therapy for treatment of suspected intrathecal baclofen withdrawal syndrome.

Key Words: Baclofen, Cyproheptadine, Withdrawal Symptoms, Rehabilitation
The value of intrathecal baclofen (ITB) therapy in spastic hypertonia of cerebral or spinal origin has been well established. Unfortunately, ITB delivery has been associated with a number of complications ranging in severity from inconvenient to life threatening. The most significant of these include pump and catheter malfunctions, withdrawal syndromes, and infection necessitating removal of hardware.

In ITB administration, one of the most concerning complications is interruption of medication administration. This interruption is a common end result of pump malfunction, catheter blockage, or catheter dissociation. Interruption of ITB administration has been shown to lead to a withdrawal syndrome. This syndrome has a wide range of severity, from increased spastic hypertonia and pruritis to seizures and audiovisual hallucinations. In its most severe form, ITB withdrawal syndrome presents a constellation of symptoms described as mimicking autonomic dysreflexia, neuroleptic–malignant syndrome, or malignant hyperthermia. The associated morbidity and mortality of ITB withdrawal syndrome has been well documented. Given the resemblance of ITB withdrawal syndrome to the above syndromes, the standard of treatment has thus far been the administration of baclofen and benzodiazepines, with some arguments for the administration of dantrolene.

The most recent development in the treatment of ITB withdrawal syndrome was proposed by Meythaler and associates, who propose the use of cyproheptadine for treatment of ITB withdrawal. Their logic was based on the similarity of ITB withdrawal syndrome to serotonergic syndromes, as in overdoses of selective serotonin reuptake inhibitors. They postulated that ITB withdrawal syndrome may actually be a form of serotonergic syndrome caused by downregulation of γ-aminobutyric acid (GABA) B receptors, and that addition of a serotonergic antagonist would prove of value. This concept led to the successful administration of cyproheptadine prophylactically in patients displaying early signs and symptoms of ITB withdrawal at the authors’ institution.

Given the increasing use of ITB therapy for spasticity in the pediatric patient, ITB withdrawal syndrome is likely to become more common, and a need for treatment of the syndrome has arisen. We present the first published use of cyproheptadine in ITB withdrawal syndrome in a pediatric patient, and we propose that its use in addition to the administration of baclofen and benzodiazepines may prove of benefit to the pediatric physiatrist who manages children receiving ITB therapy.

CASE REPORT

A 50-kg, 14-yr-old female with a history of cerebral palsy and spastic tetraplegia was previously treated successfully with ITB for 18 mos. She underwent an uneventful pump refill in an outpatient setting. Her reservoir returned less than 1 ml of baclofen, but no low-volume alarm was noted. She returned home without incident. Approximately 8 hrs later, she developed severe pruritis and increased spasticity. She was taken to the emergency room of a pediatric hospital, where she was evaluated and found to have a low-grade fever (37.8°C), tachycardia (pulse 123 beats per minute), hyperreflexia, clonus, and increased spasticity. Agitation and feelings of fear were noted. Her lower-extremity modified Ashworth scale was increased from 1+ to 3, her deep tendon reflexes were increased from 0 to 3+, and her right ankle displayed seven to eight beats of clonus, increased from two. Her left ankle displayed no clonus.

In the emergency department, her pump was interrogated and found to be functioning normally at her previously programmed dose of 520 μg/day, simple continuous administration. Radiographic evaluation of pump and catheter were without change from prior studies, with pump in place and catheter tip terminating at the superior endplate of the sixth thoracic vertebrae. She was given 20 mg of baclofen orally with 6 mg of cyproheptadine (one and one half 4-mg tablets) for a diagnosis of presumed ITB withdrawal. Symptoms abated dramatically within 1 hr. Her agitation diminished, her tachycardia and fever resolved, and her spasticity, hyperreflexia, and increased right ankle clonus likewise returned to her original baseline. After telephone consultation with technical representatives of the pump manufacturer, a diagnosis of pump stall was reached, and a 50-μg intrathecal bolus of baclofen was administered prophylactically via pump. She was admitted for overnight observation with orders for 20 mg of oral...
baclofen every 6 hrs and 6 mg of cyproheptadine every 6 hrs for symptoms of ITB withdrawal.

On the morning after admission, she was again evaluated and found to have remained afebrile overnight. Her lower-extremity spasticity was a 1+ on the modified Ashworth scale; she displayed only two beats of clonus at the right ankle. Her deep tendon reflexes could not be elicited. Her pump residual volume was changed from 2 to 3 ml to avoid recurrences of ITB withdrawal. After completing 24 hrs of observation, she was discharged home with a prescription for oral baclofen and cyproheptadine for use if necessary after telephone consultation with a physician.

**DISCUSSION**

Baclofen is a selective ligand of the bicuculline insensitive GABA<sub>B</sub> receptors present on primary afferent terminals and highly concentrated in laminae I to IV of the dorsal horn of the spinal cord. On binding, it inhibits the influx of calcium, which prevents the release of the excitatory neurotransmitters glutamate and aspartate. Receptor downregulation was shown in mouse models by Hwang and Wilcox. In humans, oral baclofen is routinely used as a skeletal muscle relaxant for spasticity. Central nervous system depression limits the maximal allowable oral dose. The concept of intrathecal administration is to deliver the medication directly to its site of action in the spinal cord, allowing for much smaller doses and minimizing side effects. Unfortunately, the introduction of ITB therapy also introduced new complications, related both to the hardware implantation and medication administration.

Intrathecal baclofen therapy has been related to both baclofen overdose and withdrawal. Overdose signs and symptoms include flaccidity, hyporeflexia, respiratory depression, apnea, seizures, coma, autonomic instability, hallucinations, hypothermia, and cardiac conduction abnormalities. Management includes supportive care and removal of as much baclofen as possible from the hardware. If not contraindicated, physostigmine may be used to reduce central side effects such as somnolence and respiratory depression.

The use of benzodiazepines and dantrolene sodium in the treatment of ITB withdrawal syndrome has been drawn from experience in the treatment of neuroleptic malignant syndrome (NMS). Both NMS and ITB withdrawal syndrome do share some similarities in presentation. The most overlap has been observed in autonomic symptoms, with fever, tachycardia, and hypotension documented in cases of each, and as observed in our patient. However, NMS is thought to be caused by dopamine blockade or reduced transmission to the thalamus. Intrathecal baclofen withdrawal syndrome, on the other hand, has been linked to serotonin syndrome. Meythaler and associates have advanced the theory that GABA inhibits the release of serotonin through GABA<sub>B</sub> receptors at the level of the brainstem. The serotonergic pathway seems to adjust to this long-term inhibition. With abrupt discontinuation of this inhibition, as in our patient, the system seems to swing back toward release of excessive serotonin. This manifests in the patient as a form of serotonin syndrome, forming the basis for the use of cyproheptadine in its treatment.

Although ITB withdrawal syndrome is perhaps the most feared complication of ITB therapy, no defined treatment algorithm exists for this entity in the pediatric population. Current pediatric treatment standards are based on experiences in the adult population and consist of administration of baclofen, either orally or intrathecally, and benzodiazepines. In our case, the use of oral cyproheptadine in conjunction with oral baclofen allowed sufficient time for pump interrogation and administration of intrathecal baclofen bolus, without progression of the patient’s withdrawal symptoms. Although it is possible that the symptom abatement was attributable entirely to the administration of oral baclofen, one would not expect such a profound response to oral baclofen in a patient receiving ITB. In addition, the cyproheptadine provided excellent subjective symptomatic relief of pruritis. Pruritis is considered a frequent but inconsistent symptom of ITB withdrawal syndrome. Although not pathognomonic, it helps differentiate between ITB withdrawal and aggravation of spasticity stemming from other causes. In our experience, the appearance of pruritis and its relief by the administration of cyproheptadine helped guide clinical decision making to proceed with intrathecal bolus of baclofen, despite a normal diagnostic interrogation of the patient’s intrathecal pump.

Our success in this case has led to a change in the standard education of the family of the ITB pump implantation patient. Previous education did not emphasize pruritis, but we now provide information regarding the symptoms of ITB withdrawal syndrome, with particular emphasis on return of spasticity and the appearance of pruritis. All patients are now discharged with prescriptions for cyproheptadine, in addition to oral baclofen. In the event of subjective increase of spasticity or pruritic symptoms, patient families are given instructions to immediately call their physician. If instructed, they administer oral baclofen and cyproheptadine and proceed to the emergency room of our institution. We continue to provide families with information to distribute to medical staff of other hospitals if they are unable to travel to our institution or are traveling themselves.
As illustrated in the case report, prompt initiation of cyproheptadine in conjunction with the standard administration of oral and intrathecal baclofen proved successful in averting the progression of ITB withdrawal syndrome in our patient. We recommend that its use in children receive further study, given the prevalence of implantable ITB pumps in this patient population.

REFERENCES


Physical Medicine and Rehabilitation Board Review: Palm Edition


This program is a digitized version of the Physical Medicine and Rehabilitation Board Review book. It allows the loading of a condensed version of the book onto a personal digital assistants (PDA). It is available for Palm devices as well as Windows-based devices.

Overall, this is a good program. It was easy to use and easy to load onto a PDA. Once on the PDA, navigation is fairly easy. The information seems accurate, although the text is hard to read on a Palm device. Palm devices with older Palm operating system software may have problems reading some of the files. The text can get distorted, and some letters may be changed to symbols. There also seem to be problems running the program off a peripheral storage source such as a memory stick or memory card.

Although the program is largely useful, some subjects could use more elaboration. For instance, the orthotics and prosthetic section would be better if a quick reference guide were added, including the K1–4 levels and what they mean, how to measure leg-length discrepancy, how to perform the Thomas test, and how to measure the popliteal angle. Pictures of the different types of sockets, knees, and feet would also be useful. In the EMG section, pictures of the different waveforms would be helpful. A chapter about interventional physiatry would also be beneficial. Wound care is an important topic that does not receive much coverage.

This program was evaluated by several residents. All agreed that it would be helpful for board review. The book version was felt to be better, but the PDA version was considered good as a supplement to be used while on the go. Their biggest complaint was that the program was hard to read because of the size of the text. Several residents were interested in purchasing the program.

Rating: ★★★★★

Lam Nguyen, DO
San Antonio, Texas
INSTRUCTIONS TO OBTAIN CATEGORY 1 CME CREDITS:

1. Read the Designated CME Articles in this issue.

2. Read the following CME Self-Assessment Exam Questions.

3. Photocopy and complete the CME Self-Assessment Exam Answering Sheet and CME Evaluation.

4. Send the completed Answering Sheet and Evaluation to: Bradley R. Johns, Managing Editor, CME Department-AAP, American Journal of Physical Medicine & Rehabilitation, 7240 Fishback Hill Lane, Indianapolis, IN 46278

CME Self Assessment Exam Questions

CME Article Number 5: Saveika, et al.

1. Baclofen is a ligand of which receptor?
   A. GABA _A_  
   B. GABA _B_  
   C. Bicuculline  
   D. Tetrodotoxin

2. \( \gamma \)-aminobutyric acid B (GABA \_B\) has been postulated to inhibit which neurotransmitter?
   A. Serotonin  
   B. Dopamine  
   C. Gutaamate  
   D. Aspartate

3. Which of the following is a symptom of intrathecal baclofen withdrawal?
   A. Pruritis  
   B. Bradycardia  
   C. Hyporeflexia  
   D. Psychomotor retardation

4. Which of the following is most likely to interrupt delivery of intrathecal baclofen?
   A. Pump malfunction  
   B. Pump migration  
   C. Systemic infection  
   D. Localized trauma

5. Treatment of intrathecal baclofen withdrawal should include all of the following EXCEPT:
   A. Baclofen  
   B. Cyproheptadine  
   C. Benzodiazepines  
   D. Clonidine

This is an adult learning experience and there is no requirement for obtaining a certain score. The objective is to have each participant learn from the total experience of studying the article, taking the exam, and being able to immediately receive feedback with the correct answers. For complete information, please see “Instructions for Obtaining Continuing Medical Education Credit” at the front of this issue.

Every question must be completed on the exam answering sheet to be eligible for CME credit. Leaving any item unanswered will make void the participant’s response. This CME activity must be completed and postmarked by December 31, 2008. The documentation received will be compiled throughout the calendar year, and once a year in January, participants will receive a certificate indicating CME credits earned for the prior year of work. This CME activity was planned and produced in accordance with the ACCME Essentials.
The answers to any essay questions must be typed or computer printed on a separate piece of paper and attached to this page.

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2. Complete the CME Evaluation and Certification on the following page and mail to Bradley R. Johns, Managing Editor, CME Dept.-AAP, American Journal of Physical Medicine & Rehabilitation, 7240 Fishback Hill Lane, Indianapolis, IN 46278.
3. This educational activity must be completed and postmarked by December 31, 2008. AAP Members may complete and submit this CME Answering Sheet and the following CME Evaluation and Certification page online through the members-only section of the AAP web page at www.physiatry.org.

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Journal Issue Month and Year ____________________________
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CME Article Author’s Name __________________________

Circle the appropriate answers.

1. A B C D
2. A B C D
3. A B C D
4. A B C D
5. A B C D
Was the article consistent with the stated objectives?  

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Did reading this article prepare you to achieve its stated objectives?  

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Is reading this article likely to enhance your professional effectiveness?  

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Was the article format conducive to learning?  

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Suggestions for future topics: ________________________________

I, _______________________________________________________, certify that I have met the criteria for CME credit by studying the designated materials, by responding to the self-assessment questions, by reviewing those parts of the article dealing with any question(s) answered incorrectly, and by referring to the supplemental materials listed in the references.

This educational activity is designated for 1½ category 1 CME credits.

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Perceived Preparedness for Physiatric Specialization and Future Career Goals of Graduating Postgraduate Year IV Residents during the 2004–2005 Academic Year

ABSTRACT


Objective: The purpose of this study was to evaluate trends among postgraduate year (PGY) IV physiatry residents, at the time of graduation from residency, in terms of their perceived experiences in the core clinical areas, confidence with procedural subspecialization, choice in career specialization, and desire to pursue clinical fellowship.

Design: Surveys were distributed to 386 PGY IV residents in physiatry at the end of the 2004–2005 academic year.

Results: Ninety-three residents (24%) completed responses in a confidential manner. Residents who were generally more confident in core clinical areas, as defined by the Self-Assessment Examination, and specialty prescription writing also believed themselves to be more prepared to practice these topics in their careers. Overall levels of confidence and perceived preparedness correlated positively with months of training and negatively with the belief in the need for postresidency fellowship training to incorporate these areas into clinical practice. Positive correlations also existed among perceived levels of preparedness in performing various physiatric procedures. Statistically significant differences in levels of confidence and preparedness existed among geographic regions when evaluating core physiatric subject matter. Fifty-six percent of residents who responded planned to pursue fellowship training, and a majority of residents intended to perform interventional procedures and musculoskeletal medicine in their practices.

Conclusions: These results provide insight into how trainees perceive their current clinical education. With validation of measures for confidence and preparedness, this survey may be useful as an adjunct resource for residency programs to evaluate their trainees.

Key Words: PM&R Residency Training, Residency Education, Resident Career Goals, Regional Residency Training, PM&R Procedures, Resident Fellowship Goals
As the field of physical medicine and rehabilitation evolves, many residency programs have noted a trend for recent graduates to pursue fellowship training and specialization. However, data evaluating physiatric residency education and trends in resident career choices is sparse. Though correlations have been shown between quartile ranking on the Self-Assessment Examination (SAE) and passing the American Board of Physical Medicine and Rehabilitation (ABPMR) Part I Board Certification Examination, it is unclear how residents perceive their current education in terms of learning the clinical core areas and preparing for future specialization. During the 1987–1988 academic year, a survey of chief residents from 70 board-accredited physical medicine and rehabilitation residency programs found great variability in the total months of residency devoted to various clinical specialties in physiatry. A 1993 survey found that trainees believed they needed more clinical and didactic experience in industrial medicine, sports medicine, and therapeutic injections. During the past few years, however, residents and recent graduates have shown interest in musculoskeletal medicine, soft-tissue disorders, therapeutic injections, and nerve blocks. This study was conducted to evaluate trends among postgraduate year (PGY) IV physiatry residents, at the time of graduation from residency, in terms of their perceived experiences in the core clinical areas, confidence with procedural subspecialization, choice in career specialization, and desire to pursue clinical fellowship.

**METHODOLOGY**

Physiatric residency programs accredited by the Accreditation Council for Graduate Medical Education (ACGME) were identified using the ACGME Web page (www.acgme.org) in March 2005. On the basis of information provided, each program coordinator was sent a package in May 2005 and was asked to distribute the contents among graduating PGY IV residents in their respective program. Enclosed for each resident were an introductory letter, a questionnaire, and a postage-paid, preaddressed unmarked envelope to protect confidentiality. Residents were asked to return the surveys by mail within 4 wks of receiving them.

The questionnaire was designed to address clinical areas as outlined by the SAE and established by the ABPMR for the 2004–2005 academic year (Table 1). In the first section, residents were asked to rate their levels of confidence in evaluating physiatric diagnoses, and their perceived levels of preparedness to enter a career in specific areas. The response options were 1 = very unsure/very unprepared, 2 = somewhat unsure/somewhat unprepared, 3 = neutral, 4 = somewhat confident/somewhat prepared, and 5 = very confident/very prepared. They were asked about the need for fellowship training to continue a career path after residency (1 = yes, 0 = no), and the number of months of training devoted to specific disciplines during their residency. The second section addressed perceived levels of preparedness for performing specialized procedures (Table 1). The third section consisted of open-ended questions to determine plans for fellowship training and career goals. Finally, residents rated their confidence levels in recommending workplace modifications, prescribing prosthetic or orthotic devices, and prescribing physiatric therapeutics. Though residents did not provide identifying markers to protect confidentiality, they were asked to provide the name of their current residency training program to help categorize data according to geographic regions (East Coast, Midwest, South, and West Coast). Using the Statistical Package for the Social Sciences (SPSS) computer software, the data were analyzed for (a) general descriptive information, (b) relationships among various factors, and (c) differences among geographic region.

**RESULTS**

Of 386 surveys distributed, 93 responses were returned, yielding an overall response rate of 24%,
TABLE 2  Response rate by geographic region

<table>
<thead>
<tr>
<th>Region</th>
<th>Contribution to Study, %</th>
<th>Number of Responses</th>
<th>Total Possible Responses</th>
<th>Response Rate, %</th>
</tr>
</thead>
<tbody>
<tr>
<td>East</td>
<td>41.9</td>
<td>39</td>
<td>170</td>
<td>23</td>
</tr>
<tr>
<td>South</td>
<td>28.0</td>
<td>26</td>
<td>76</td>
<td>34</td>
</tr>
<tr>
<td>Midwest</td>
<td>14.0</td>
<td>13</td>
<td>95</td>
<td>14</td>
</tr>
<tr>
<td>West</td>
<td>16.1</td>
<td>15</td>
<td>45</td>
<td>33</td>
</tr>
<tr>
<td>Totals</td>
<td>100</td>
<td>93</td>
<td>386</td>
<td>24</td>
</tr>
</tbody>
</table>

with ranges of 14%–34% per region (Table 2). Ninety-one residents responded that they had spent their entire residency within the same program, one resident responded that he or she had not, and one resident provided no response. Overall, there were significant correlations between: preparedness and confidence ($r = 0.983; P < 0.001$); confidence and months of training within specific clinical areas ($r = 0.844; P < 0.001$); preparedness and months of training ($r = 0.849; P < 0.001$); and preparedness and need for fellowship training ($r = −0.655; P < 0.03$).

Respondents were most confident and believed themselves to be most prepared in the areas of musculoskeletal medicine and electrodiagnosis (Table 3). They were least confident and prepared in pediatric rehabilitation. Correlations ranging from 0.726 to 0.936 ($P < 0.001$) existed between levels of confidence and preparedness in all core rehabilitation specialties. Negative associations with belief in a need for fellowship training were noted for both levels of confidence ($r = −0.216$ to $−0.553; P < 0.04$) and levels of preparedness ($r = −0.209$ to $−0.518; P < 0.05$) in specific areas, except that results for medical rehabilitation were not statistically significant. For most core rehabilitation specialties, significant relationships existed between levels of confidence and total months of training ($r = 0.268−0.471; P < 0.02$); however, results for brain disorders, electrodiagnostics, and industrial rehabilitation were not statistically significant. Significant relationship also existed between levels of preparedness and total months of training ($r = 0.246−0.519; P < 0.01$), except that results for electrodiagnostics were not significant.

With regard to writing physiatric therapeutic prescriptions, creating prosthetic and orthotic (P&O) prescriptions, and recommending workplace modifications, there were associations between levels of confidence in the core clinical areas ($r = 0.547−0.839; P < 0.001$) and levels of preparedness for specialty prescription writing in that clinical area ($r = 0.514−0.915; P < 0.001$; Table 4). Months of training correlated positively with confidence in writing P&O prescriptions and prescriptions for physiatric therapeutics ($r = 0.428$ and $0.463$, respectively; $P < 0.001$). Confidence in writing P&O prescriptions and recommending workplace modifications were negatively related to belief in the need for fellowship training in the respective core specialty ($r = −0.388$ and $−0.291$, respectively; $P < 0.01$).

Residents who believed they were better prepared in performing acupuncture also believed they were better prepared in performing interventional physiatric procedures ($r = 0.277; P = 0.007$) and osteopathic manual manipulation ($r = 0.301; P = 0.003$). Resident levels of perceived preparedness in both interventional physiatry and periph-

TABLE 3  Mean responses regarding core clinical areas

<table>
<thead>
<tr>
<th>Clinical Area</th>
<th>Confidence</th>
<th>Preparedness</th>
<th>Belief for Need for Fellowship, %</th>
<th>Months Overall of Training</th>
</tr>
</thead>
<tbody>
<tr>
<td>Brain disorders</td>
<td>3.98</td>
<td>3.72</td>
<td>32</td>
<td>4.61</td>
</tr>
<tr>
<td>Electrodiagnostics</td>
<td>4.28</td>
<td>4.18</td>
<td>14</td>
<td>6.16</td>
</tr>
<tr>
<td>Industrial rehabilitation</td>
<td>3.45</td>
<td>3.35</td>
<td>24</td>
<td>2.39</td>
</tr>
<tr>
<td>Joint and connective tissue disorders</td>
<td>3.69</td>
<td>3.52</td>
<td>23</td>
<td>3.40</td>
</tr>
<tr>
<td>Medical rehabilitation</td>
<td>3.83</td>
<td>3.79</td>
<td>11</td>
<td>5.42</td>
</tr>
<tr>
<td>Musculoskeletal</td>
<td>4.41</td>
<td>4.23</td>
<td>23</td>
<td>7.22</td>
</tr>
<tr>
<td>Neuromuscular disorders</td>
<td>3.51</td>
<td>3.35</td>
<td>39</td>
<td>3.96</td>
</tr>
<tr>
<td>Pediatric rehabilitation</td>
<td>3.19</td>
<td>3.02</td>
<td>73</td>
<td>2.77</td>
</tr>
<tr>
<td>Physiatric therapeutics</td>
<td>4.14</td>
<td>4.11</td>
<td>3</td>
<td>5.16</td>
</tr>
<tr>
<td>Prosthetics/orthotics</td>
<td>3.67</td>
<td>3.59</td>
<td>12</td>
<td>3.32</td>
</tr>
<tr>
<td>Spinal cord injury</td>
<td>4.16</td>
<td>3.96</td>
<td>38</td>
<td>3.91</td>
</tr>
</tbody>
</table>
eral joint aspiration/injections were significantly correlated with preparedness in performing phenol and/or botulinum toxin injections ($r = 0.256$ and $0.359$, respectively; $P < 0.02$). Finally, a relationship was noted between levels of preparedness for interventional physiatry and peripheral joint aspiration/injections ($r = 0.375; P < 0.001$).

Analyses of variances revealed statistically significant geographic differences in confidence levels and preparedness levels for certain subject matter. Specifically, East Coast residents were more confident than West Coast residents in joint and connective tissue disorder (means: East Coast: 3.92; West Coast: 3.13; $P = 0.004$). Southern residents were more confident than West Coast residents in prescribing P&O (means: South: 3.92; West Coast: 3.07; $P = 0.01$), and Southern residents were more confident than East Coast residents in spinal cord injury medicine (means: South: 4.60; East Coast: 3.76; $P = 0.001$). Residents in both the South and East Coast believed they were more prepared in prescribing P&O than did West Coast residents (means: South: 3.96; East Coast: 3.62; West Coast: 2.87; $P < 0.03$). Southern residents believed they were more prepared than did East Coast residents (means: South: 4.36; East Coast: 3.59; $P = 0.006$) in spinal cord injury medicine.

When asked to note future career paths, most residents indicated that they planned to pursue interventional procedures and musculoskeletal medicine (Table 5). Fifty-two respondents (56%) indicated that they planned to pursue a fellowship after graduation from residency. Of the residents surveyed, 95.3% of those pursuing careers in interventional procedures practice (significance: two sided, $<0.001$, with forward exact test) planned to pursue fellowships to continue their training.

**DISCUSSION**

The results of this study indicate that resident confidence and preparedness generally correlated with total months of training in core physiatric disciplines and specialty prescription writing. Trainees who were more confident also believed themselves to be better prepared to enter careers in those clinical areas, and those individuals who believed they were better prepared generally felt that postresidency fellowship training was not necessary for professional clinical work.

Surveys of confidence and preparedness have been used by various medical specialties to evaluate physician competence and clinical experience. A longitudinal cohort study conducted in 2000 measured overall preparedness of pediatric residents in core clinical areas, and evaluated these same individuals after 5 years of clinical practice. In general, it was found that the residents’ perceptions of preparedness at the time of graduation were similar to retrospective assessments 5 years later. With respect to subspecialties in which there were differences in perceived preparedness, practicing physicians felt they may have been more prepared than they initially thought they were at the time of residency completion. Similarly, a 2005 survey of recently graduated obstetric–gynecology residents found that overall levels of confidence in managing clinical problems remained constant after graduation in core areas, although it was noted that periodic review of graduates’ perception of training could be used to adjust residency curriculum relative to future clinical needs.

**TABLE 4** Mean perceived preparedness for specialty prescriptions and procedures

<table>
<thead>
<tr>
<th>Specialty Areas</th>
<th>Average Preparedness</th>
</tr>
</thead>
<tbody>
<tr>
<td>Physiatric therapeutic prescriptions</td>
<td>4.04</td>
</tr>
<tr>
<td>Peripheral joint injections</td>
<td>4.00</td>
</tr>
<tr>
<td>Botox/phenol injections</td>
<td>4.00</td>
</tr>
<tr>
<td>Prosthetic/orthotics prescriptions</td>
<td>3.52</td>
</tr>
<tr>
<td>Workplace modification prescriptions</td>
<td>3.28</td>
</tr>
<tr>
<td>Epidural injections and nerve blocks</td>
<td>2.70</td>
</tr>
<tr>
<td>OMT</td>
<td>2.19</td>
</tr>
<tr>
<td>Acupuncture</td>
<td>1.61</td>
</tr>
</tbody>
</table>

**TABLE 5** Career paths and interests of graduates in 2004–2005

<table>
<thead>
<tr>
<th>Career Paths and Interests</th>
<th>Graduates Planned to Pursue</th>
<th>Number of Responses</th>
<th>Percentage of Respondents</th>
</tr>
</thead>
<tbody>
<tr>
<td>Interventional procedures practice</td>
<td></td>
<td>43</td>
<td>46</td>
</tr>
<tr>
<td>Musculoskeletal practice</td>
<td></td>
<td>32</td>
<td>34</td>
</tr>
<tr>
<td>General practice</td>
<td></td>
<td>17</td>
<td>18</td>
</tr>
<tr>
<td>Outpatient practice</td>
<td></td>
<td>9</td>
<td>9.7</td>
</tr>
<tr>
<td>Sports practice</td>
<td></td>
<td>9</td>
<td>9.7</td>
</tr>
<tr>
<td>Inpatient practice</td>
<td></td>
<td>6</td>
<td>6.5</td>
</tr>
<tr>
<td>Spinal cord injury practice</td>
<td></td>
<td>6</td>
<td>6.5</td>
</tr>
<tr>
<td>Electromyography practice</td>
<td></td>
<td>4</td>
<td>4.3</td>
</tr>
<tr>
<td>Traumatic brain injury/stroke practice</td>
<td></td>
<td>2</td>
<td>2.2</td>
</tr>
<tr>
<td>Private practice</td>
<td></td>
<td>1</td>
<td>1.1</td>
</tr>
</tbody>
</table>

*The sum of the results is greater than 100% because respondents had multiple responses.*
implemented to evaluate specific areas during family practice residency, and have even been used as an internal gauge for these residencies to evaluate their overall effectiveness in teaching ACGME core competencies to their trainees. Finally, though resident perception of comfort is multifactorial, a 2006 survey of internal medicine residents found that self-perceived levels of comfort in performing clinical procedures was associated with the number of procedures performed. Despite well-documented criteria required by the ACGME for clinical education of residents, it has been noted that some difficulty exists in assessing and measuring overall resident competence, and identifying appropriate methods of evaluation for residency programs. The most recent program requirements for residency education established by the residency review committee of the ACGME state that residents should have the following clinical experiences before graduation: at least 12 mos of direct responsibility for the complete management of inpatients; at least 12 mos in the care of outpatients, with significant emphasis on patients with musculoskeletal disorders; and approximately 200 electromyographic consultations. Beyond these requirements, residency programs have a great deal of the flexibility to design their curricula when incorporating the clinical competencies required for a broadly based and complete physiatric clinical exposure. This flexibility may account for the great variability between residency programs in designing their clinical and didactic schedules.

In addition to the SAE, scheduled resident and program director meetings, and annual program internal reviews, the measures used in this study may provide a supplemental resource to help monitor the progress of trainees. With further studies, correlations may be shown between resident self perception and success on board certification or subspecialty certification examinations. Quantitative analysis could also be conducted to determine the appropriate number of patient encounters or months of clinical exposure, as they relate to reaching appropriate confidence and preparedness levels for professional success. As an adjunct method of evaluation, these data may assist residency programs when designing clinical and didactic components to focus on the core rehabilitation disciplines and newly evolving clinical and procedure-based experiences that trainees will need for future subspecialization.

Similar analyses could be used to evaluate the procedure-based curriculum in residency programs. This study indicates that there are positive correlations between perceived preparedness levels among different physiatric procedures. It is unclear whether perceived preparedness in one procedural area tends to generalize to perceived preparedness in other procedural areas or whether individuals with an interest in procedural based medicine seek out these procedures. However, according to the data collected relevant to career paths, procedure-based practice will play an important role in the clinical work of future physiatrists. Further analysis of resident exposure to these procedures may help determine optimal exposure during residency to provide clinical competence after graduation. Furthermore, such information may provide further insight as to what types of procedures will yield the most effective results in preparing a resident for clinical practice. This seems to be of particular importance given the percentage of residents from this study that plan to incorporate procedures in their future work.

Residency education and experiences may significantly differ according to the prevalence of certain clinical diagnoses within various geographic areas. If a residency program’s clinical experience is biased secondary to its geography, it may not have appropriate resources to provide exposure in other facets of physiatry. This, in turn, could account for variable levels of confidence and preparedness between regions. With further geographic information, residency programs may be able to actively adjust clinical experiences to provide a well-rounded physiatric education.

The questions posed in this survey yield interesting results regarding resident self perception of clinical skills, time devoted to clinical areas during residency education, and career and fellowship plans for graduating residents. However before generalizing the results of this study, potential sources of error must be examined. A major threat to the external validity of this study was the relatively low response rate of 24%. Because of the potential for response bias, caution must be used when interpreting the data. More than 40% of the respondents to this survey stated they planned to pursue interventional physiatry practice after graduation, and more than 30% planned to pursue musculoskeletal medicine. It is possible that residents with these career plans were more likely to respond to the survey. There is also potential bias for the results of this study to be skewed according to the viewpoints and experiences of individuals pursuing interventional, as well as musculoskeletal, based practice. Similar considerations for bias must be made when evaluating the geographic data in this study, because a majority of respondents hailed from the East Coast and South. Though producing graduates with increased beliefs of competence may be a desired outcome of residency education, confidence and perceived preparedness in this study have not been validated with more objective measures, such as the SAE or ABPMR Part I Certification Examination.
tion, months of training are traditionally used by residency programs to determine adequate resident exposure to clinical areas of training, but it is not clear whether months of training, vs. specific patient encounters, is the appropriate method to quantitatively evaluate clinical experience. Finally, the use of open-ended questions to describe fellowship plans and career choices yielded a diverse array of responses. A possible source of error was the subsequent interpretation and assignment of these responses into more traditional categorizations.

To counter the issue of low response rate, a repeat study should be conducted to further increase both regional and national response rates. With the assistance and cooperation of residency program directors, graduating residents could complete the survey either in paper form or with an Internet-based questionnaire. The survey could be administered at the time of the resident exit interviews for graduation to ensure that the data collected reflect the most comprehensive evaluation of experiences. With confidential coding of individual residents completing the surveys, correlations could then be established between their subjective answers of perceived confidence and preparedness with objective scores on either the SAE or ABPMR Part I Certification Examination. Furthermore, the use of a more detailed survey with closed-ended questions could eliminate the ambiguity of open-ended responses regarding future career plans.

In conclusion, this survey presents interesting data pertinent to the evaluation of resident experiences and career choices. Future studies are needed with larger sample sizes to generalize results to the overall graduating resident population, specifically with respect to overall levels of confidence and preparedness in various physiatric specialties, geographic differences between resident perceptions, and future career plans. With validation of confidence and preparedness measures, similar studies may provide residency programs an additional resource to evaluate their trainees.

REFERENCES

Outcome Assessment in Randomized Controlled Trials of Stroke Rehabilitation

ABSTRACT


The lack of a unified approach to outcome assessment in stroke rehabilitation limits our ability to interpret evidence provided by randomized controlled trials (RCTs). The purpose of this review was to identify outcomes and assessment tools reported in RCTs of stroke rehabilitation interventions as a first step toward consistent assessment of outcomes. Given that the validity of research is linked to reliability and validity of measurement, the relationship between the use of previously developed outcome measures and the methodological quality of RCTs was explored.

Electronic literature searches identified RCTs examining stroke rehabilitation therapies from 1968 to 2005. The Physiotherapy Evidence Database (PEDro) scale was used to assess methodological quality. Cited outcomes were recorded and assessment tools identified as previously published or study specific. Four hundred ninety-one RCTs cited the assessment of 1447 outcomes using 489 measurement tools. Two hundred fifty-four of these were previously published, and 235 were study specific. A core of 30 frequently cited tools was identified. The use of previously published assessment tools to evaluate primary study outcomes was associated with higher PEDro scores.

Significant heterogeneity in outcome assessment was demonstrated, although a core of 30 frequently cited tools could be identified. Appropriate evaluation and selection of outcome measures would enhance the methodological quality of randomized controlled trials.

Key Words: Outcome Assessment, Evidence-Based Medicine, Reliability, Validity, Stroke Rehabilitation, Measurement, Randomized Controlled Trials
Stroke has a broad impact on individuals, families, and the communities in which they live. Numerous assessment tools have been created to assess the complex and heterogeneous consequences of stroke and the effect of stroke rehabilitation interventions on stroke outcomes. However, there is no consensus regarding what and how outcomes should be measured to create a comprehensive description of individual functioning and health and to facilitate our ability to make comparisons across interventions and studies. The current lack of a unified approach to assessment of outcome limits our ability to interpret the evidence provided by randomized controlled trials (RCTs) in a clinically meaningful way.

Our ability to interpret study results is also limited by the validity of the research itself, which, in turn, is limited by the reliability of outcome assessment. Ideally, researchers should use, whenever possible, measurement tools that have been previously evaluated for use in similar subject populations and have demonstrated acceptable reliability, validity, and responsiveness. The revised CONSORT guidelines recommend the use of previously developed and validated tools to improve quality of measurement and facilitate between study comparisons.

As a first step toward developing consensus around consistent assessment of stroke rehabilitation outcomes, an examination of the outcome assessment tools that have been reported in the stroke rehabilitation literature was conducted. Given that the interpretability and utility of research results is associated with the validity of outcome measurement, the association between the use of previously developed outcome measures and the methodological quality of RCTs was examined.

METHODS

Literature Search

The present study arose from the Evidence-Based Review of Stroke Rehabilitation. Search strategies included the following databases: MEDLINE, CINAHL, EMBASE, MANTIS, PASCAL, Psych-Info and Sci Search from 1968 to 2005. The literature search, study selection, and data abstraction strategies employed for that review process have been described in detail elsewhere. For the present study, randomized controlled trials (RCTs) examining primary or secondary stroke prevention treatments were excluded, as were review articles, meta-analyses, or systematic reviews. RCTs were included if they cited the use of a measurement tool or scale to assess at least one of the study outcomes. Studies assessing only discrete, counted outcomes (e.g., mortality, length of stay, occurrence of a stroke event, etc.) were excluded. On the basis of these criteria, 491 RCTs evaluating the effectiveness of interventions in stroke rehabilitation were selected for inclusion.

Data Abstraction and Quality Assessment

Two abstractors, each blinded to the other’s results, reviewed each article independently. The review process consisted of two parts. In the first part, data pertinent to the study were collected, including study methodology, cited outcomes, and means by which study outcomes were assessed. After this, the study was assigned a quality-rating score derived using the Physiotherapy Evidence Database (PEDro) scale.

The PEDro scale, developed by the Centre for Evidence-Based Physiotherapy (CEBP) in Australia (www.pedro.fhs.uwyd.edu.au), was created to assess bibliographic details and abstracts of RCTs, quasi-randomized studies, and systematic reviews in physiotherapy. Based on a set of core criteria for quality assessment in randomized controlled trials derived by expert consensus, the scale consists of ten items (Table 1). Each item receives a rating of either yes (1) or no (0). Item ratings are

<table>
<thead>
<tr>
<th>TABLE 1</th>
<th>The Physiotherapy Evidence Database (PEDro) scale</th>
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<tbody>
<tr>
<td>1.</td>
<td>Subjects were randomly allocated to groups (in a crossover study, subjects were randomly allocated an order in which treatments were received)</td>
</tr>
<tr>
<td>2.</td>
<td>Allocation was concealed</td>
</tr>
<tr>
<td>3.</td>
<td>Groups were similar at baseline regarding the most important prognostic indicators</td>
</tr>
<tr>
<td>4.</td>
<td>There was blinding of all subjects</td>
</tr>
<tr>
<td>5.</td>
<td>There was blinding of all therapists who administered the therapy</td>
</tr>
<tr>
<td>6.</td>
<td>There was blinding of all assessors who measured at least one key outcome</td>
</tr>
<tr>
<td>7.</td>
<td>Adequacy of follow-up (could all of the subjects originally randomized be accounted for at the end of the study?)</td>
</tr>
<tr>
<td>8.</td>
<td>Intention-to-treat analysis</td>
</tr>
<tr>
<td>9.</td>
<td>Results of between-group statistical comparisons were reported for at least one outcome</td>
</tr>
<tr>
<td>10.</td>
<td>Study provides both point measures and measures of variability for at least one key outcome</td>
</tr>
</tbody>
</table>

Eligibility criteria were included in the original PEDro scale. Because subject selection pertains more directly to external rather than internal study validity, it was not included in the present review.
summed to provide a total score. Unlike checklists such as the Downs and Black,13 the PEDro scale does not include evaluation of study outcomes and, therefore, provides an evaluation of methodological quality unbiased by the selection of outcome measures. Four of the PEDro items have been empirically validated (randomization, concealed allocation, blinding, and adequacy of follow-up), and the remaining items have face validity.11 Reported intraclass correlation coefficients for interrater reliability for the PEDro scale range from 0.5614 to 0.91.15 A moderate correlation (r = 0.59) between PEDro scores and ratings of methodological quality using the Jadad scale has also been reported.16 Two independent raters, blinded to each other, assigned a PEDro score to each study. A third reviewer resolved any scoring disagreements. A detailed description of this application of the PEDro scale has been published previously.9,10

For each study included in the review, all cited outcomes were recorded. Primary outcomes were identified. For our purposes, an outcome was considered to be primary if it was explicitly named as the primary outcome by the study author(s) or if it was listed first in the study methodology. Each measurement tool cited was categorized as previously published or study specific. Tools were classified as previously published if prior publication of the tool along with published, evaluation(s) of reliability or validity or responsiveness could be identified from the information provided by the study authors. Study-specific tools were those developed for the project at hand; they had no previously published evidence of reliability, validity, and/or responsiveness cited by the study authors.

Mean (± SD) and median (interquartile range) PEDro scores were calculated for studies citing previously published outcome measures and also for studies citing the use of study-specific tools. In addition, mean and median PEDro scores were calculated for studies using previously published or study-specific tools to assess the primary study outcome. Given that the PEDro scale yields nonparametric data, median scores for studies using previously published measures were compared with those citing the use of study-specific measures using the Mann–Whitney U test (a nonparametric alternative to the t test). An alpha level of <0.05 was considered to be statistically significant. All statistical calculations were performed using SPSS for Windows (version 12.0).

RESULTS

One thousand seven hundred twenty-one outcome citations were recorded from 491 identified RCTs. Of these, 274 represented assessments of clinical variables, which were described but made no reference to the use of a specific measurement scale. The remaining 1447 cited study outcomes were assessed using 489 assessment tools. Of these, 254 had been previously published, and 235 (48.1%) were study specific.

Of the 254 previously published scales, only 30 were cited more than ten times each. These 30 most frequently cited measurement tools accounted for approximately 50.1% (725) of all outcome citations. A list of the most frequently cited assessment instruments is provided in Table 2.

One hundred fifty-two studies cited the use of one or more study-specific measures to assess outcomes. Of these, 66 cited the use of a study-specific measure to assess the primary study outcome. Mean (± SD) and median PEDro scores as well as mean ranks are provided in Table 3.

Although the median PEDro score in the group of studies including study-specific assessment tools is the same as the studies using only

<table>
<thead>
<tr>
<th>TABLE 2</th>
<th>Frequently cited outcome measures</th>
</tr>
</thead>
<tbody>
<tr>
<td>Most Frequently Cited Outcomes 1968–2005</td>
<td>No. of Citations</td>
</tr>
<tr>
<td>Barthel index</td>
<td>123</td>
</tr>
<tr>
<td>Timed walk assessments (varying times and distances)</td>
<td>72</td>
</tr>
<tr>
<td>Fugl–Meyer assessment of stroke recovery</td>
<td>58</td>
</tr>
<tr>
<td>Modified ashworth scale</td>
<td>43</td>
</tr>
<tr>
<td>Functional independence measure</td>
<td>38</td>
</tr>
<tr>
<td>Mini-mental state examination</td>
<td>27</td>
</tr>
<tr>
<td>Action research arm test</td>
<td>25</td>
</tr>
<tr>
<td>Frenchay activities inventory</td>
<td>22</td>
</tr>
<tr>
<td>Nottingham health profile</td>
<td>22</td>
</tr>
<tr>
<td>Medical outcomes study short form</td>
<td>22</td>
</tr>
<tr>
<td>Nottingham extended activities of daily living</td>
<td>21</td>
</tr>
<tr>
<td>Motor assessment scale</td>
<td>21</td>
</tr>
<tr>
<td>Hospital anxiety and depression scale</td>
<td>17</td>
</tr>
<tr>
<td>Berg balance scale</td>
<td>16</td>
</tr>
<tr>
<td>Rankin handicap/modified Rankin/Oxford handicap</td>
<td>16</td>
</tr>
<tr>
<td>Nine-hole peg test</td>
<td>15</td>
</tr>
<tr>
<td>Hamilton rating scale for depression</td>
<td>15</td>
</tr>
<tr>
<td>Motricity index</td>
<td>14</td>
</tr>
<tr>
<td>General health questionnaire</td>
<td>14</td>
</tr>
<tr>
<td>Rivermead mobility inventory</td>
<td>13</td>
</tr>
<tr>
<td>Rivermead motor assessment</td>
<td>13</td>
</tr>
<tr>
<td>VAS–pain</td>
<td>12</td>
</tr>
<tr>
<td>Porch index of communicative ability</td>
<td>12</td>
</tr>
<tr>
<td>Motor activity log</td>
<td>12</td>
</tr>
<tr>
<td>Brunnstrom scale</td>
<td>11</td>
</tr>
<tr>
<td>Caregiver strain index</td>
<td>11</td>
</tr>
<tr>
<td>Timed up and go</td>
<td>10</td>
</tr>
<tr>
<td>Functional ambulation categories</td>
<td>10</td>
</tr>
<tr>
<td>Scandinavian stroke scale</td>
<td>10</td>
</tr>
<tr>
<td>Weschler memory subtests</td>
<td>10</td>
</tr>
</tbody>
</table>
Previously published measures, both the mean PEDro scores and mean rank reflect a significant difference in distribution toward lower PEDro scores in the group using study-specific measures ($z = -2.51, P = 0.012$). Among the subset of studies citing the use of study-specific measures, those studies using a previously published measure to evaluate the primary study outcome obtained a higher median and mean PEDro score than did the group of studies using a study-specific measure for this purpose. Use of a previously published measure to assess primary study outcomes was significantly associated with higher PEDro scores ($z = -2.6, P = 0.009$).

**DISCUSSION**

As in previous examinations of RCTs in stroke research, considerable heterogeneity of measurement was noted. Four hundred ninety-one RCTs cited the use of 489 measurement tools in the assessment of 1447 study outcomes. This variability and inconsistency in the selection and assessment of outcomes limits our ability to interpret individual study data and make appropriate comparisons between trials of similar interventions. However, a core of 30 frequently cited, previously published assessment tools was identified. These tools were used in the assessment of a wide range of patient outcomes along the continuum from body structure and function assessments to health-related quality of life and could, together with rigorous examination of their psychometric properties, provide a starting point around which a consensus for outcome assessment in stroke rehabilitation trials could begin.

Despite the availability of a wide selection of previously published measures, almost one half (48%) of all measurement tools identified in the present review were study specific. Marshall et al. have identified the use of unpublished rating scales as a major source of bias in randomized controlled trials of nonpharmacological treatments of schizophrenia, such that trials using unpublished scales were significantly more likely to report significant effects in favor of treatment when compared with control conditions. The authors suggest several possible explanations for why unpublished scales could be a source of bias: (i) unpublished scales tend to be used by small trials of poorer quality, (ii) selective reporting of data obtained from unpublished scales, eliminating nonsignificant results, or (iii) ad hoc adjustment to the content of unpublished scale to present the most favorable data. Although this bias has not been demonstrated in other branches of medicine, it is recommended that investigators refrain from using unpublished assessment tools. Indeed, it is generally accepted that reliable and valid measurement is essential in the production of meaningful and replicable data, and, to that end, researchers should endeavor to use measures that are known to be reliable and valid.

The present study compared the methodological quality of trials citing the use of previously published measures with those using study-specific tools. Although use of a previously published measure, particularly in the assessment of the primary study outcome, was associated with superior methodological quality, the difference between groups was approximately equivalent to a single point on the PEDro scale. Total PEDro scores provide an indication of which trials are more likely to be valid and contain sufficient statistical information to enhance interpretability. A difference of a single point may represent an important deficiency in methodology, particularly if the item were considered to be a significant source of bias, such as concealed allocation, blinding of patients or therapists, or intention-to-treat analysis.

The apparent association between the use of previously published measurement tools and the methodological quality of RCTs signals the importance of outcome assessment to research. The selection of outcome measures may be as important a component to the overall quality of an RCT as are methods for blocking, concealed allocation, adequacy of follow-up, and other components of research design. The selection of appropriate, reliable, and valid measurement tools should be more

<table>
<thead>
<tr>
<th>TABLE 3 Mean and median Physiotherapy Evidence Database (PEDro) scores for studies using previously published vs. study-specific measures</th>
</tr>
</thead>
<tbody>
<tr>
<td>All studies ($n = 491$)</td>
</tr>
<tr>
<td>Cited only previously published measures ($n = 339$)</td>
</tr>
<tr>
<td>Included study-specific measures ($n = 152$)</td>
</tr>
<tr>
<td>Studies including study-specific measures ($n = 152$)</td>
</tr>
<tr>
<td>Used previously published measure to assess primary outcome ($n = 86$)</td>
</tr>
<tr>
<td>Used study-specific measure to assess primary outcome ($n = 66$)</td>
</tr>
</tbody>
</table>
formally recognized and consistently reported in a standardized manner, as recommended by Dijkers et al. and Moher et al.

Limitations of the Present Study

In this study, previously published outcomes were recorded separately from and compared with study-specific measures. Previously published measures were defined, for the purposes of the present paper, as those that had been published previously and had published evidence available with regard to reliability, validity or responsiveness. However, no specific claims can be made regarding the measurement properties of an assessment tool based solely on prior publication within the research literature. Indeed, confirming whether these previous publications provide evidence of adequate reliability, validity, or responsiveness, or whether previous evaluations of the psychometric or clinimetric properties of a given assessment tool had been conducted within a population of individuals who had experienced stroke, was beyond the scope of the present review. Similarly, with regard to the tools designated as study specific, the present review neither reported whether reliability/validity data were presented nor evaluated whether any such data were appropriate or acceptable.

CONCLUSIONS

Our understanding of the effectiveness of a given rehabilitation intervention is, in part, determined by our ability to measure the impact of a given intervention on an individual’s functional ability and health. However, there is no consensus regarding what and how outcomes should be measured. This study identified significant heterogeneity of outcome assessment in a review of 491 randomized controlled trials of stroke rehabilitation interventions. Although there were literally hundreds of measures used in the assessment of outcomes, a core of 30 well-known measures accounted for more than one half of the measurement citations found in RCTs examining the efficacy of stroke rehabilitation interventions.

Standardized evaluation of outcome measures, and the process by which measures are selected for use, would undoubtedly enhance the methodological quality of RCTs in stroke rehabilitation research. It would also improve the confidence with which research results can be interpreted, reproduced, and applied.

ACKNOWLEDGMENTS

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Persistent Hiccups During Rehabilitation Hospitalization
Three Case Reports and Review of the Literature

ABSTRACT


Persistent hiccups have been reported to occur occasionally during rehabilitation hospitalizations. Hiccups can interfere with patient participation and progress, and this can lengthen and complicate the rehabilitation hospitalization. Chlorpromazine, the only Food and Drug Administration–approved agent specifically indicated for hiccups, is often chosen as a first-line treatment. However, chlorpromazine does not always provide favorable results.

The intent of this paper is to (1) review hiccups and implications of persistent hiccups in the rehabilitation setting, (2) provide additional evidence that chlorpromazine is often not the best treatment choice for hiccups in rehabilitative patients, (3) report the effectiveness and favorable tolerability of modest doses of gabapentin in a small case series of three patients, and (4) review potentially effective treatment approaches for hiccups.

Key Words: Hiccups, Hiccoughs, Singultus, Gabapentin, Stroke, Postoperative

OVERVIEW OF HICCUPS

Hiccups\(^1\,^2\) (also hiccoughs or singultus) involve sudden, abrupt inspiration resulting from sudden, involuntary contraction of the diaphragm. Closure of the glottis then halts the incoming air. The column of air strikes the closed glottis to produce the characteristic sound. Hiccups most often involve unilateral contraction of the diaphragm (the left more frequently than the right).

The reflex arc for hiccups includes an afferent limb (including the vagus and phrenic nerves and the sympathetic chain), a central mediator, and an efferent limb (including the phrenic nerve, plus efferents to the glottis and inspiratory intercostal muscles). The reflex pathways are similar to those that produce coughing, sneezing, swallowing, and vomiting.

Hiccups do not seem to have any physiologic purpose or function after birth, but speculation exists that intrauterine hiccups permit training of the diaphragm without aspiration of amniotic fluid. Most cases of hiccups are of short duration, lasting less than 48 hrs. No gender difference seems to exist in the occurrence of transient hiccups. Brief bouts of hiccups do not require medical intervention.
Transient hiccups are benign in nature and occur nearly universally in all individuals from time to time. Such events are often spontaneous but can be related to gastric overdistention (such as overeating, drinking carbonated beverages, aerophagia, and gastric insufflation during endoscopy), sudden changes in ambient or gastrointestinal temperature, alcohol use, tobacco use, sudden excitement, or emotional distress.

Intractable hiccups occur more frequently in men, and about 4000 hospitalizations per year occur in the United States for hiccups. The longer the duration of hiccups, the more likely an organic cause exists. Hiccups occurring at night also have a greater chance of having an organic vs. a psychogenic cause. Numerous potential causes of medically significant hiccups have been described, most commonly including central nervous system disorders, lesions of the vagus and/or phrenic nerve, toxic–metabolic disorders, adverse events associated with pharmacological agents, and psychological disorders. Hiccups are also not uncommon in the postoperative state.

The longer hiccups persist, the more difficult treatment becomes. Persistent hiccups can prolong hospitalizations by contributing to debilitation with diminished nutritional and fluid intake, aspiration, insomnia, mental stress, and even wound dehiscence.

REVIEW OF THE REHABILITATION LITERATURE

Two published articles concerning hiccups were identified in the rehabilitation literature. Nickerson et al. report that hiccups developed in a 69-yr-old male with lateral medullary syndrome during his acute care stay. He was given chlorpromazine, promethazine, and prochlorperazine, without success. On admission to rehabilitation, his main complaints were of hiccups at a rate of 69/min, associated with nausea. At the rehabilita-

tion facility, baclofen (5 mg orally, three times a day) was substituted for the promazine, with prompt resolution of hiccups, diz-

ziness, precluding participation in rehabilitative therapies. The above reports highlight that hiccups can occur in individuals with stroke usually affecting the brainstem. Complications can occur and can protract the rehabilitation length of stay. Chlorpromazine was poorly tolerated, and various other agents (baclofen, haloperidol, and carbamazepine) provided resolution of the hiccups.

CASE REPORTS

Three cases are reported in which persistent hiccups interfered with rehabilitation program participation and in which prompt hiccup resolution was achieved with the use of gabapentin.

Case 1

A 60-yr-old male was admitted to a medical unit after the acute onset of ataxia, dizziness, and decreased coordination in the right upper extremity. Hiccups at a rate of 10–15/min began 15–20 mins after onset of the neurological symptoms. A magnetic resonance imaging scan showed the presence of a lateral medullary infarct. The patient was admitted to rehabilitation 9 days after onset, still with hiccups. His hiccups caused persistent nausea, associated with emesis, with resultant poor fluid and nutritional intake. Blood urea nitrogen and creatinine had increased from 16 and 0.9 mg/dl at time of acute care admission to 47 and 1.5 mg/dl, respectively, during the sixth day of his stay on the rehabilitation unit. Baclofen was initially used while on the medical unit at a dose of 5 mg orally three times a day, then increased to 10 mg orally three times a day on the rehabilitation unit, without success. Chlorpromazine at a dosage of 25 mg orally three times a day was substituted for the baclofen. The patient’s hiccups improved, but he developed worsening dizziness and orthostatic hypotension, precluding participation in rehabilitation therapies. Gabapentin (100 mg orally, four times a day) was then substituted for the chlorpromazine, with prompt resolution of hiccups, dizziness, nausea, and emesis, and normalization of laboratory studies; blood urea nitrogen and creatinine values had decreased to 15 and 1.0 mg/dl, respectively, at a follow-up visit 3 wks later.

Comments on Case 1

Hiccups previously have been described as associated with lateral medullary infarction (also known as posterior inferior cerebellar syndrome, or Wallenberg syndrome). Park et al. present a series of 51 patients with lateral medullary infarction; magnetic resonance imaging scanning within...
3 days of onset confirmed the diagnosis. Seven of these 51 patients developed hiccups in addition to the more typical symptoms of vertigo, dizziness, nausea, vomiting, and dysphagia.

Pelin et al. describe four patients with lateral medullary syndrome who also presented with hiccups. These patients were treated with varied doses and combinations of chlorpromazine, valproic acid, and baclofen. Full resolution of hiccups was observed in all patients.

Moretti et al. describe the use of gabapentin in 15 patients (nine men and six women; mean age, 69.1 yrs) who experienced hiccups caused by lateral medullary infarcts. The authors report success in all of the patients with use of gabapentin, as was the case with our patient. Two years after a stroke, one patient had a recurrence of hiccups that again responded to gabapentin. Follow-up neurologic examination in this patient remained stable, and repeat magnetic resonance imaging demonstrated the previous stroke, but no acute findings were detected. Moretti described the following gabapentin protocol: 400 mg three times a day for 3 days, then 400 mg daily for 3 days, and then discontinuation of gabapentin. No significant adverse effects were reported from the gabapentin.

Case 2

A 72-yr-old female was initially seen with complaints of headache and clumsiness in the right lower extremity. She was brought to the emergency department. Computed tomographic scan of the head was negative. Concerns were expressed about the upper cervical spine (possible calcification vs. cavernous angioma vs. small area of hemorrhage). She was scheduled for a follow-up clinic appointment.

Four days later, she developed increased weakness in the right upper and lower extremities, and she fell in her bathroom. She was brought to the emergency department and was admitted. Coagulation studies showed an international normalized ratio of 3.5.

Her past history was significant for hypertension, breast cancer, paroxysmal atrial fibrillation, and remote deep venous thrombosis with pulmonary embolism 20 yrs previously (after bilateral mastectomy for breast cancer). She had been on chronic anticoagulation with warfarin, and a Greenfield filter had been placed previously.

Computed tomography of the cervical spine soon after admission demonstrated a hematoma localized to the right side of the cervical spinal cord (C2 and above), extending to the pontomedullary junction. Anticoagulation was reversed with fresh-frozen plasma. After normalization of her international normalized ratio, she was admitted to rehabilitation, with persistent right hemiparesis. Cervical collar was used to stabilize her cervical spine because of the presence of the hematoma and because she complained of dizziness with just minimal neck movements. She slowly, steadily improved to the point where she was able to become independent with ambulation and self-care activities. She was discharged home to live alone after a 22-day rehabilitation unit stay. Follow-up computed tomographic scan of the cervical spine before discharge demonstrated nearly complete resolution of the hematoma.

Interestingly, at the time of admission to rehabilitation, the patient indicated that she was experiencing persistent hiccups occurring at a frequency of two to three per minute; she reported that the hiccups had been present for 3 days and had begun about 4 days after the worsening of her right hemiparesis. She stated that the hiccups were annoying to her and that they interfered with eating and drinking activities. She was prescribed chlorpromazine (25 mg orally, three times a day), and the hiccups greatly improved; however, she complained of dizziness, and she developed orthostatic hypotension, which precluded participation in her rehabilitation program. The chlorpromazine was discontinued, and gabapentin (100 mg orally, four times a day) was initiated, with total resolution of the hiccups and dizziness. The gabapentin was tapered and discontinued without any recurrence of hiccups.

Comments on Case 2

Hemorrhage within the spinal canal is a rare but serious complication of anticoagulation therapy, described most often in individuals in the sixth or seventh decade of life. Most cases reported in the literature seem to be spontaneous, but others are described as secondary to trauma (epidural catheters, spinal manipulation, snowboarding, nose blowing), small-cell lung cancer, metastatic hepatocellular cancer, or coagulopathies. Patients described in many of the reported cases had been anticoagulated within the therapeutic range. The diagnosis should be considered if a patient taking anticoagulant medication complains of local or referred spinal pain and has associated limb weakness, sensory deficits, or urinary retention. Anticoagulation should be reversed promptly. Emergency laminectomy and decompression of the spinal cord are often recommended (Pullarkat et al. describe these measures as “mandatory”), but cases in which the patient is already improving rapidly and steadily and is under close observation could be treated nonoperatively.

Hemorrhage adjacent to the spinal canal is sufficiently rare; only limited numbers of cases have been described in the literature. This is the first case reported in which hiccups were present in association with such a hemorrhage. The location...
of the hemorrhage—from the pontomedullary junction descending to C2—is similar in location to the lesions seen with lateral medullary stroke, implicating similar neurological factors in producing the hiccups.

**Case 3**

A 65-yr-old type 2 diabetic male was admitted to rehabilitation 6 days after bilateral total-knee arthroplasties. After surgery, he had redness and swelling around his left leg that were thought to be attributable to cellulitis. He had been maintained on deep venous thrombosis prophylaxis. Doppler ultrasound studies for deep venous thrombosis were negative. The cellulitis was treated with cephalexin, with improvement in the redness, swelling, and pain. His overall rehabilitation course was slow because of pain and decreased active knee motion. Hydrocodone was used for pain control. He also had hypoalbuminemia (2.6 g/dl) and required psychosocial encouragement and supplements to boost his nutrition.

The patient developed hiccups (3–5/min), which persisted during his last day on the surgical service and for his first day on rehabilitation; the hiccups aggravated his postsurgical pain. Gabapentin (100 mg orally, three times a day) was initiated, with prompt resolution of the hiccups. After 8 days on the rehabilitation unit, he had improved to the point where he was ready for hospital discharge with outpatient therapy. Hiccups did not recur.

**Comments on Case 3**

Souadjian and Cain analyzed 220 patients with intractable hiccups seen at the Mayo Clinic. Risks produced by intractable hiccups were reported as severe debilitation, dehydration, insomnia, protracted mental stress, wound dehiscence, and, possibly, even death. Eighty-two percent of the patients were male. Forty patients (18% of the total group) were reported to have postsurgical hiccups. Hiccups usually appeared 1–4 days postoperatively and were generally severe and interfered with sleep. Initial abortive measures failed in 65% of cases, and pharmacological measures were usually required to resolve the hiccups.

In our patient, an elderly diabetic male, gabapentin was chosen as the initial treatment for his hiccups. He had a prompt, complete resolution of hiccups on low-dose gabapentin, and he tolerated this medication without adverse effects.

**REVIEW OF TREATMENT APPROACHES FOR PERSISTENT HICCUPS**

A variety of folk remedies and physical maneuvers have been described as helpful in aborting hiccups (see Table 1). These measures seem to be most effective in transient hiccups and are less effective with persistent hiccups.

<table>
<thead>
<tr>
<th>TABLE 1 Folk remedies and other nonpharmacological treatments of hiccups</th>
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<tbody>
<tr>
<td>Breathing into a bag</td>
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<tr>
<td>Drinking from opposite side of glass</td>
</tr>
<tr>
<td>Swallowing granulated sugar</td>
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<tr>
<td>Drinking or gargling ice water</td>
</tr>
<tr>
<td>Forcible traction on tongue</td>
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<tr>
<td>Bitting on a lemon</td>
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<tr>
<td>Eating a spoonful of peanut butter</td>
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<tr>
<td>Compression of the eyeballs</td>
</tr>
<tr>
<td>Carotid massage</td>
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<tr>
<td>Rectal massage</td>
</tr>
<tr>
<td>Catheter or swab stimulation of the naso-oropharynx</td>
</tr>
<tr>
<td>Valsalva maneuver</td>
</tr>
<tr>
<td>Breath holding</td>
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<tr>
<td>Fright</td>
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<tr>
<td>Noxious odors (inhaling ammonia)</td>
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<tr>
<td>Gastric lavage</td>
</tr>
<tr>
<td>Modified Heimlich maneuver. Three moderate-pressure thrusts at 10-sec intervals</td>
</tr>
<tr>
<td>Sexual intercourse</td>
</tr>
</tbody>
</table>

A careful history, thorough physical examination, and studies, as appropriate, should be performed to help identify conditions or disorders that might be producing or perpetuating the hiccups. The list of potential causes of hiccups is long, including central and peripheral neurological lesions, head and neck lesions, cardiothoracic and abdominal disorders, infectious diseases, toxic/metabolic causes, postoperative status, and psychogenic causes.

For persistent hiccups, the patient should be questioned about any medication that might be producing or aggravating the hiccups. Medication cessation or a drug “holiday” should be considered (if possible) if a potentially offending medication is identified. A number of different medications have been implicated in producing or aggravating hiccups (see Table 2).

Review of treatment options should take into consideration the potential precipitating or aggravating causes of the hiccups. For example, if hiccups are related to gastroesophageal reflux disease, a proton pump inhibitor might be quite effective in relieving symptoms.

Often, no obvious cause is identified to account for the hiccups. In these cases, pharmacologic treatment is often needed. Initially, monotherapy is recommended, but occasionally a combination of agents is required to treat persistent hiccups.

Chlorpromazine is the only medication specifically approved by the Food and Drug Administration for the treatment of hiccups. Use for hiccups was first described by Friedgood and Ripstein in
be effective. More recent reports indicate that for times a day, for 7–10 days) also has been shown to prevent the development of hypotension. Oral chlorpromazine (25–50 mg, three to four hours to prevent the development of hypotension. Chlorpromazine, if chosen for elderly patients. Chlorpromazine, if chosen for patients had a more profound response to the medication. The dose was, therefore, decreased in elderly patients. Chlorpromazine, if chosen for intravenous use, must be infused slowly for several hours to prevent the development of hypotension. Oral chlorpromazine (25–50 mg, three to four times a day, for 7–10 days) also has been shown to be effective. More recent reports indicate that for elderly and potentially neurologically impaired rehabilitation patients, chlorpromazine is best avoided because of the problematic adverse effects of dizziness with orthostatic hypotension and drowsiness, which can preclude participation in active rehabilitation programs. Two of the patients in this study experienced adverse responses to chlorpromazine, providing further evidence that chlorpromazine is often not the most appropriate treatment choice for hiccups in rehabilitative patients.

Numerous other medications have been reported as effective for treating hiccups. Omeprazole has been found helpful in patients in whom the hiccups seem to be related to the presence of gastroesophageal reflux disease. Baclofen and anticonvulsants (phenytoin, valproic acid, carbamazepine, and gabapentin) also can be effective in some cases. Occasionally, in resistant cases of hiccups, combinations of these medications are necessary. Other treatment approaches for resistant cases include hypnosis, acupuncture, cervical epidural block, phrenic nerve block, crushing or pacing, and general anesthesia with positive pressure ventilation and muscle relaxants.

Gabapentin was found to be well tolerated and symptomatically useful for our three patients. Gabapentin is a Food and Drug Administration–approved anticonvulsant that is also frequently used for neuropathic pain, including diabetic neuropathic pain and postherpetic neuralgic pain. The specific anticonvulsant mechanism of action is unclear; the agent is structurally related to gamma-aminobutyric acid (GABA), but it does not interact with GABA receptors, it is not metabolically converted into GABA or a GABA agonist, and it is not an inhibitor of GABA uptake or degradation. Somnolence, dizziness, ataxia, and fatigue are the most frequent adverse effects.

Hernandez et al. report a case of a 71-yr-old male with Guillain–Barre syndrome and metastatic gastric neoplasm who presented with intractable hiccups. The neoplasm had infiltrated the anterior pillar of the diaphragm at the time of surgery. Omeprazole (20 mg daily) was initiated along with chlorpromazine (25 mg, three times a day). Hiccups, as well as emesis, persisted. Chlorpromazine was discontinued and baclofen was added, initially 5 mg three times a day, and progressively increasing to 15 mg three times a day. The patient still failed to improve. Gabapentin was then added at a dosage of 300 mg three times a day. Within 24 hrs, the hiccups ceased. One week later, hiccups recurred and resolved within 48 hrs after resumption of gabapentin at a dosage of 400 mg daily.

Porzio et al. describe gabapentin as being useful in treating hiccups in patients with cancer. A 63-yr-old male with metastatic liver cancer developed chronic hiccups, nausea, and fatigue. Metclopramide and dexamethasone prescribed by his primary care physician were ineffective. Chlorpromazine (25 mg intravenously, twice daily) eased the hiccups but caused postural hypotension and severe drowsiness. Chlorpromazine was discontinued, and gabapentin (300 mg, three times daily) was initiated. The gabapentin was well tolerated and provided effective relief from the hiccups. Two other cases—a 43-yr-old male with pancreatic cancer, and a 51-yr-old male with metastatic small-cell lung cancer—were reported to obtain relief of persistent hiccups with 300–400 mg of gabapentin three times a day. The authors caution about the use of baclofen in elderly patients because of the frequent adverse effects of sedation, insomnia, dizziness, weakness, ataxia, and confusion.
SUMMARY

Most cases of hiccups are transient and benign. Home treatments are initially reasonable and are often effective. The longer hiccups persist, the more resistant to treatment they become. Intractable hiccups can be associated with a number of pathologic conditions. A detailed history and physical examination with appropriate testing should be performed on patients who are experiencing intractable hiccups.

Chlorpromazine, despite Food and Drug Administration approval for hiccups, is often poorly tolerated and is best avoided for older rehabilitative patients, especially those with debility, acute stroke, or neurological events. Baclofen can be helpful in promoting hiccup resolution, but it should be used cautiously in elderly patients because of the potential adverse effects of sedation, insomnia, dizziness, weakness, ataxia, and confusion.

Gabapentin at modest doses, in this case series and others, has been found to provide prompt relief of hiccups, and it is usually well tolerated. A short course often can provide long-term relief. Measures combining pharmacotherapy with more invasive treatment are occasionally indicated for more resistant cases of hiccups.

REFERENCES
Persistent Hiccup Associated with Intrathecal Morphine Infusion Pump Therapy

ABSTRACT

Intraspinal drug-delivery therapy has been increasingly used in patients with intractable nonmalignant pain syndromes during the past two decades. Morphine, the only FDA-approved opioid for intrathecal administration, has been the principle agent for such therapy. Although intrathecal morphine infusion can produce profound spinal analgesia, it may also cause some untoward side effects. We describe the first case of persistent hiccup caused by intrathecal morphine infusion therapy.

Key Words: Hiccup, Intrathecal Opioid Infusion Pump, Intrathecal Morphine, Intrathecal Hydromorphone

Hiccup (singultu, hiccough) is a common phenomenon that involves sudden inspiration, immediately followed by active closure of glottis.¹ It is an involuntary, reflex-like activity that begins with contraction of the diaphragm, quickly terminated by the abrupt closure of the glottis. Hiccups are experienced at one time or another by almost everyone and, when transient, are usually harmless. Despite a common occurrence, the cause, mechanism, and significance of hiccups are still incompletely understood.² Although the causes are often considered idiopathic, more than 100 known organic causes have been identified.³ The most common causes are gastrointestinal, such as gastric distention or gastroesophageal reflux disease.⁴ Metabolic derangements, as well as drugs, are also frequently implicated as probable causes.¹,⁴

We report the first case of intractable hiccups presumably caused by intrathecal morphine infusion.

CASE REPORT
A 65-yr-old female, referred to our multidisciplinary pain center, with a 5-yr history of severe low-back pain and left-sided leg pain from degenerative lumbar disc disease, lumbar spinal stenosis, and lumbar radiculitis. The patient also had a history of lumbar laminectomy with decompressive fusion at L4–L5 and L5–S1, about 2 yrs ago after she had undergone multimodality treatment, including medication trials, physical therapy, and spinal interventional treat-
ment, including epidural steroid injections, lumbarosacral transformaminal steroid injections, lumbar facet blocks, and lumbar facet radiofrequency rhizotomy, with only short-term efficacy. The patient described her low-back pain as being about as bothersome as her left-leg pain. Her pain level was usually at 8–9/10 on a numeric pain scale of 0–10. Her low-back pain generally worsened by sitting and standing, whereas her left-leg pain, which she described as starting from the posterior thigh, extending down to the posterior calf, and then to the bottom of her left foot, significantly worsened by walking. The only relieving factor was frequently changing positions. She had no significant past medical history or surgical history, except for lumbarosacral decompressive fusion about 2 yrs before presenting to our clinic. Her family history and review of systems were noncontributory. Her medications included extended-release morphine (30 mg, thrice daily), oxycodone/acetaminophen (10/325, thrice daily, as needed), gabapentin (800 mg, thrice daily), tizanidine (4 mg, twice daily), and meloxicam (7.5 mg, twice daily). She had been on these drugs for more than 6 mos. Her lumbar MRI with and without contrast revealed broad-based bulging at L3–L4, with degenerative changes of the facet joints and bilateral neuroforaminal narrowing, more so on the left side. Postsurgical changes were at L4–L5 and L5–S1 with bilateral laminectomy defects and compression plates with interpedicular screws at L4, L5, and S1.

Because she had failed all other conventional treatment, we considered her for intraspinal drug-delivery therapy. After a psychological evaluation confirming her candidacy, she underwent an outpatient controlled continuous epidural morphine trial. A tunneled lumbar epidural catheter was placed at L3–L4, with the catheter tip advanced to L1 under fluoroscopic guidance. Satisfactory catheter placement was confirmed by epiduragram. The proximal tip of the catheter was then tunneled subcutaneously and was connected to a Microject PCEA pump (Codman, Raynham, MA) and a reservoir bag containing 0.4 mg/ml of preservative-free morphine. The pump was programmed to deliver a basal rate of 0.5 ml/hr. The bolus dose was 0.2 ml, with a 60-min lockout interval. The patient was then instructed how to use the pump and was discharged home. During the trial, the infusion rate was further increased to 1 ml/hr, with her bolus dose increased to 0.4 ml. The infusion trial lasted 7 days and was beneficial in controlling her pain. The patient reported more than 80% pain reduction, with improved distance for ambulation. She did experience initial, transient itching, which completely resolved. She did not feel the need to use any extra boluses while on epidural morphine infusion at 0.4 mg/hr (0.4 mg/ml at 1 ml/hr). She did not experience any hiccup during this epidural morphine-infusion trial. She subsequently consented and was scheduled for permanent intrathecal morphine pump implantation. The intrathecal catheter was inserted at the left paramedian L2–L3, with the catheter tip located at T12, as confirmed under fluoroscopy. A nonprogrammable Codman 3000 continuous-rate infusion pump was placed in the right-middle quadrant below the right rib cage and above the right iliac crest. The intrathecal infusion consisted of 1.0 mg/day of preservative-free morphine.

The patient started experiencing hiccups about 12 hrs after the intrathecal pump implantation. She did experience satisfactory analgesia of her usual back and leg pain, and there was no other intolerable side effect, except for the hiccups. These occurred at a frequency of four to six per minute. The patient was instructed to try different maneuvers, including holding her breath, drinking water, etc., without efficacy. The patient then tried chlorpromazine as needed, which did help her slightly. She was able to sleep at night, and her hiccups became less forceful. She also noted that the interval between episodes of hiccups was increased. Yet, she still experienced episodic hiccups while taking chlorpromazine. Six days after the initial implantation, the decision was made to change her intrathecal infusion from morphine to hydromorphone at 0.3 mg/day. Her hiccups resolved completely 16 hrs after intrathecal morphine had been changed to hydromorphone. She has not experienced recurrence of her hiccups since switching to hydromorphone. She has been on intrathecal hydromorphone for almost 3 yrs now. Her current intrathecal hydromorphone-infusion rate is 2.5 mg/day.

**DISCUSSION**

The discovery of highly specific opioid receptors in the central nervous system, especially the spinal cord in 1970s, made it possible for spinal application of opioids to obtain spinally mediated antinociception. Intraspinal drug-delivery therapy has been used increasingly since the 1980s in patients with intractable nonmalignant pain who have failed to respond to conventional treatment or who could not tolerate systemic opioid therapy because of side effects. By infusing small amounts of opioid directly into cerebrospinal fluid in close proximity to the receptor sites in the spinal cord, one can achieve spinally mediated analgesia, sparing some of the side effects caused by systemic opioids. In our own practice, most of our patients who ended up requiring intrathecal opioid infusion pumps for better pain control were those with intractable low-back pain and/or leg pain from failed back surgery syndrome, with/or without adhesive arachnoiditis. Morphine, the only FDA-ap-
proved opioid for intrathecal administration, has been found to be associated with numerous side effects. The most common ones are pruritus, nausea and vomiting, urinary retention, water retention, constipation, and sexual dysfunction. Less common ones include mental status change, central nervous system excitation, hyperalgesia, neurotoxicity, and respiratory depression. To the best of our knowledge, hiccups induced by intrathecal morphine infusion have never been reported. No record of any publications in English on hiccups associated with intrathecal opioid use could be found after Medline search using hiccup(s) and intrathecal opioid(s). Hydromorphone is considered a safe, alternative opioid for long-term intrathecal use when morphine fails because of pharmacological side effects or inadequate pain relief. Systemic opioids have been implicated in causing hiccups, although there is a paucity of data in the English literature. Wilcox has reported one case of a 63-yr-old man with severe neck pain from squamous cell carcinoma of right tonsil, experiencing persistent hiccups after taking slow-released oral morphine. Hiccups resolved only after cessation of oral morphine, with no recurrent hiccups after substitution with a fentanyl patch and oxycodone. Throughout the period when he was experiencing hiccups, the patient had no other apparent side effects that would suggest an opioid overdose. Lauterbach has reported a case of a 55-yr-old man who experienced persistent hiccups for 4 days after receiving hydrocodone (5 mg, every 4 hrs) after dental procedures, resolving after hydrocodone cessation. Bagheri et al. have shown that opioids accounted for 6% of the 53 cases of drug-induced hiccups in France between 1985 and 1997. However, because of the small sample size, 6% of 53 cases would correlate to a total of three cases in 12 yrs in France.

Transient hiccups are common and usually harmless. Hiccups lasting more than 48 hrs are considered persistent; those lasting more than 2 mos are considered intractable. Persistent or intractable hiccups can lead to fatigue, sleep disturbance, dehydration, and wound dehiscence in the postoperative period.

The exact mechanisms of hiccups are still not completely understood. It is believed that the hiccup reflex consists of an afferent and efferent limb. The afferent limb comprises the phrenic and vagal nerves as well as the sympathetic chain arising from thoracic segments T6–T12. The central connection is thought to involve interactions among the brainstem; the midbrain, including the respiratory center, phrenic nerve nuclei, medullary reticular formation, and hypothalamus; and unidentified areas in the cervical spinal cord between C3 and C5. Stimulation of either the afferent or efferent limb of the reflex arc triggers hiccups.

Multiple causes have been attributed to induce hiccups. The most commonly recognized ones are gastrointestinal, such as gastric distention or gastroesophageal reflex disease. Multiple pharmacological agents, including barbiturates, benzodiazepines, methyldopa, and steroids, have been reported to cause hiccups. More than 100 known organic causes of hiccups have been identified.

Local anesthetics given epidurally have been implicated as a cause of hiccups. McAllister et al. report one case of recurrent persistent hiccups after epidural steroid and bupivacaine injections in a 65-yr-old man with lumbar spinal stenosis. Epidural bupivacaine, rather than the steroid, was felt to be the culprit for causing hiccups, because hiccups occurred on three separate occasions after lumbar epidural injections with bupivacaine and triamcinolone, whereas hiccups did not occur after a fourth lumbar epidural injection with the same amount of triamcinolone, with normal saline instead of bupivacaine. Interestingly, to the contrary, intravenous and nebulizer lidocaine have been used as a therapeutic modality for persistent or intractable hiccups. Lower thoracic or high lumbar epidural local anesthetics may cause sympathetic blockade, resulting in contracted bowel attributable to parasympathetic dominance. However, this seems to be in contrast to the gastric distention that is thought to be frequently implicated in causing hiccups. Diaphragmatic function can also be affected by epidural blockade. Increased electrical diaphragmatic activity was noted with thoracic epidural anesthesia for upper abdominal or thoracic surgery.

The most likely explanation for thoracic epidural anesthesia-related increases in diaphragmatic activity seems to be the disruption of the inhibitory reflex of phrenic nerve motor drive, caused by either direct deafferentation of visceral sensory pathways or by diaphragmatic load reduction as a result of increased abdominal compliance.

Slipman et al. report a case of persistent hiccups associated with transforaminal thoracic epidural steroid injection with 3 ml of betamethasone and 1 ml of 1‰ lidocaine in separate occasions in a 31-yr-old man with thoracic discogenic pain. The hiccups lasted 15 and 18 hrs, respectively, and were unresponsive to conventional maneuvers, including drinking water, holding breath, and initial chlorpromazine (10 mg). The hiccups were finally aborted after four doses of metoclopramide (10 mg, every 6 hrs).

The authors attribute the hiccups to the epidural steroid, rather than lidocaine, because they believe that the injectate flow was confirmed in the epidural space, dorsal to the vertebral body, and that it did not, therefore, reach the sympathetic spinal cord.
chain that lies along the anterolateral aspects of T6–T12 vertebral bodies. However, lower thoracic or high lumbar epidural local anesthetics may cause sympathetic blockade, simply because sympathetic blockade may still occur with epidural local anesthetic, but at more a proximal anatomic location instead of at the sympathetic chain.

In our patient, hiccups occurred only after intrathecal morphine infusion with no local anesthetic infusion. She had been on oral morphine (extended-release morphine, 30 mg, thrice daily) and oxycodone/acetaminophen (10 mg, thrice daily, as needed) for more than 6 mos without experiencing any hiccups. The 1-wk, epidural morphine-infusion trial went well, with good analgesia and without any hiccups or other intolerable side effects. The hiccups persisted for 6 days despite treatment, only to resolve after the patient was switched to an intrathecal infusion of hydromorphone. It is not clear why intrathecal morphine induces hiccups, whereas intrathecal hydromorphone does not. Indeed, it is puzzling because hydromorphone, a hydrogenated ketone analog, shares similar properties to morphine, except for being more lipophilic than morphine and about five to seven times more potent than morphine. Because of ethical concerns, we did not replace intrathecal morphine infusion with normal saline and rechallenge the patient later on with intrathecal morphine to further confirm causality of hiccups by intrathecal morphine infusion.

In summary, we present a rare case of persistent hiccups after intrathecal morphine pump placement. The exact mechanism of intrathecal morphine, not hydromorphone, inducing hiccups needs further investigation. It is important to recognize this possible adverse event so that proper treatment can be initiated.

REFERENCES


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ABSTRACT


The recent case of Ashley X has sparked much recent public debate and controversy, and raises critical questions for physiatrists and rehabilitation professionals. The case came to light when Gunther and Diekema published an article in the October 2006 issue of Archives of Pediatric and Adolescent Medicine describing a novel growth attenuation treatment for Ashley X, a 6-yr-old girl with developmental disabilities. Her parents also published a blog about Ashley, with detailed explanations for their rationale in choosing this treatment on behalf of Ashley which involved beliefs about her future and quality of life. Ashley’s parents refer to the series of interventions as the Ashley Treatment in their blog, and this phrase has also been adopted by the popular press and others who have commented on the case. In this article we present an analysis of the Ashley X treatment and use a disability ethics approach to examine the perspectives of various stakeholders involved, including Ashley and other girls with extensive disabilities, parents, physicians, and bioethics committees. We conclude with critical questions for physiatrists and other disability specialists who are in a unique position to examine medical controversies involving people with disabilities.

Key Words: Disability, Ethics, Evidence-Based Medicine, Pediatrics

Recently the parents of a young girl with disabilities sought controversial medical interventions to attenuate her growth.1 This case, described by Gunther and Diekema2 in an article published in the October 2006 issue of Archives of Pediatric and Adolescent Medicine, touches on many concerns central to the practice of rehabilitation medicine. The following commentary on the Ashley X treatment uses a disability ethics approach3 to examine the perspectives of various stakeholders involved. Such an approach opens up alternative ways to frame the issues, various treatment options, and considerations for the role of health care providers as moral agents facilitating difficult decisions on behalf of children with significant cognitive and developmental disabilities.

BACKGROUND

According to her physicians, Ashley X is a disabled girl with a diagnosis of static encephalopathy, the etiology of which is unknown.2 She was born at full term after an unremarkable pregnancy and uncomplicated birth. She has two healthy siblings and lives with her parents, both of whom have college educa-
tions. She is described as being dependent in all activities of daily living, and completely immobile, hence the parents’ term “Pillow Angel”—that is, she stays where they put her. She is nonverbal and receives all of her nutrition via a feeding tube. Ashley X responds to others in her environment via smiles and vocalizations. Her developmental stage is described as that of a 3-mo-old infant, and her physicians have indicated that they do not expect this to change.

When Ashley X was 6 yrs 7 mos old, and beginning to show signs of early puberty, her parents approached a pediatric endocrinologist for an evaluation. In the 6 mos before the evaluation with the endocrinologist she had advanced from the 50th to the 75th percentile for length. The parents “particularly feared that continued growth eventually would make it untenable for them to care for their daughter at home, despite their strong desire to do so . . . After extensive consultation between parents and physician, a plan was devised to attenuate growth by using high-dose estrogen.”

To avoid complications from high-dose estrogen, Ashley also underwent a hysterectomy and breast bud removal. It is this combination of interventions that the parents later refer to as the Ashley Treatment.

The parents’ blog provides additional information about their rationale and states that Ashley requires all the care of a baby. The parents felt that they were approaching the limits of their ability to lift Ashley at 65 lbs. Other than her parents, the grandmothers were her only care providers and they were beginning to have difficulty lifting Ashley. They felt that by accelerating the closure of her growth plates it would be easier to care for her in the home and would improve her quality of life by facilitating mobility as well as community participation. Ashley’s parents were also concerned about the potential for sexual abuse if she were to develop secondary sex characteristics, discomfort from menstruation, large or fibrocystic breasts (which were common in her family) and difficulty with wheelchair harness fitting. A number of other secondary benefits and concerns were raised including the prevention of skin breakdown, infections, and ease in bathing. Quoting from the parents’ blog, they also state that “given Ashley’s mental age a nine and a half year old body is more appropriate and more dignified than a fully grown female body.”

DISABILITY ETHICS

Disability ethics incorporates a disability studies approach to traditional clinical medical ethics. Disability studies is an emerging interdisciplinary field that is grounded in the lived experience of disability and in the social model of disability. Simplistically stated, the medical model of disability characterizes disability as a defect intrinsic to the individual. The primary goal in the medical model is to cure or normalize the person when possible, to treat disease and relieve pain. In the social model, disability is seen as inherently neutral, and a product of the interactions between the person and the environment. Neither model fully explains all aspects of disability. More nuanced models see disability as a “complex interaction of biological, psychological, cultural and sociopolitical factors which cannot be extricated except with imprecision.”

In the analysis of the Ashley Treatment that follows, we have a clear illustration of just how complex these interactions are and the challenges they pose for healthcare providers.

Ethical Issues for Stakeholders: Ashley and Other Young Girls with Cognitive and Physical Disabilities

In December 2006, the United Nations General Assembly adopted the Convention on the Rights of Persons with Disabilities, stating that “every person with disabilities has a right to respect for his or her physical and mental integrity on an equal basis with others.” The American College of Obstetrics and Gynecology (ACOG) came to a similar conclusion in their position paper on the “Sterilization of women, including those with mental disabilities.” They concluded that hysterectomy should not be used as the primary and initial treatment for dealing with menstruation, or for nonvoluntary sterilization for girls with disabilities. “In disabled women with limited functional capability, indications for major surgical procedures remain the same as in other patients. In all cases, indications for surgery must meet standard criteria, and the benefits of the procedure must exceed known procedural risks.” The concerns raised by caregivers for Ashley regarding menstrual cramps and hygiene issues are only speculative. It is critical to assess if there are less risky and reversible treatments available that might be equally effective before embracing riskier and permanent interventions.

ACOG further notes that using medical treatments for social or quality-of-life reasons can be problematic. Many of the concerns raised in support of the Ashley Treatment—such as Ashley’s future care, risk of sexual abuse, wheelchair fitting, and community participation—cannot be resolved simply by medical efforts to keep her small, by performing an hysterectomy or mastectomy. On the other hand, physicians are often in a position to make referrals to other specialists (e.g., independent living centers, rehabilitation professionals, etc.) when it is clear that the problems identified extend beyond medical and biological factors to
environmental and social concerns. The ACOG and UN perspectives can both be summarized as a right to bodily integrity for people with cognitive and developmental disabilities. Involuntary (against the person’s wishes) or nonvoluntary (for those such as Ashley who cannot participate in the decision making) medical interventions and surgical procedures in general should only be provided when there are clear health benefits.

Furthermore, people with cognitive and developmental disabilities have been harmed in the past by controversial and experimental medical treatments and need added protections. The history of medicine is replete with examples of harm to people with disabilities by well-intentioned doctors, using popular but misguided and inadequately tested treatments such as prefrontal lobotomies. New treatments for those with disabilities must undergo the same scrutiny and rigorous research to elucidate the actual benefits and risks as do new medical interventions for the general population. Many people with cognitive and developmental disabilities are unable to provide informed consent and rely on others to make decisions on their behalf. They are particularly vulnerable and therefore require additional safeguards.

Ashley and other children with cognitive and developmental disabilities have interests and needs apart from those of their parents. It is difficult to separate Ashley’s interests from those of her parents—clearly her future well-being will largely be determined by the ability of her family to continue to provide or arrange for a loving home and 24-hr care. Children with developmental disabilities who are dependent upon their parents for all of their daily care and who may never be able to communicate their opinions and desires are often not seen as having distinct interests. Yet, it is precisely because of such dependencies and vulnerability that health care professionals need to be aware that society has placed some parameters around the types of decisions parents can and cannot make. For example, it is no longer acceptable that parents of newborns with Down syndrome can refuse life-saving surgeries for treatable esophageal atresias or heart anomalies, or for parents to refuse blood transfusions or high benefit life-saving interventions for their children based upon the parental religious beliefs (e.g., Jehovah’s Witnesses, or Christian Science). There are also limits in place regarding parental decisions for pediatric sterilization including state laws such as the one in Washington where Ashley had her surgery. In rare instances, the state is empowered to step in and take over the care of a child under the parens patriae power when the parents or guardians are unable or unwilling to act in the best interest of the child.

Finally, being described as a girl with the mental age of an infant may unnecessarily limit expectations for the future and ignores Ashley’s capacity for social and emotional development. Developmental skills and abilities of children with pervasive needs, in the absence of known progressive neurological disorders, can change with time, nurturance and stimulation, just as they can change for children without disabilities.

Ethical Issues for Stakeholders: Ashley’s Parents and Other Caregiving Family Members

The lack of adequate social supports, including reliable attendant care, quality respite services and affordable community-based living programs for people with disabilities remains problematic. Parents would not need to seek risky medical interventions to minimize difficulties with caregiving if they were offered reasonable alternatives. If Ashley’s parents were convinced that she would always receive loving and capable care, either in their home or in alternative community-based programs, regardless of her size and sexual maturation, their concerns could have been mitigated. This appalling lack of support lies at the center of this debate and cannot be ignored or fixed with medical interventions such as surgery and drugs.

It is also true that parental caregivers have been harmed by judgmental health care professionals who have incomplete appreciation of their lived experience as caregivers. There has been a tendency in medicine to criticize and pathologize family members who seek solutions to caregiving challenges. Parents are often viewed as selfish if they institutionalize their children, yet overprotective if they keep them at home without outside assistance. Parents are also often left with inadequate resources and information when making decisions on behalf of their children. When analyzing the treatment options for Ashley, rehabilitation professionals need to be careful not to contribute to this pattern by blaming her parents for their efforts to seek unconventional solutions in the absence of quality resources for families of children with disabilities.

It is also important to note that in most instances parents are in the best position to make decisions on their child’s behalf. Parents are imbued with the legal and moral responsibility to care for their children. In making medical decisions, they are expected to use a “best interest” standard, weighing the proposed benefits and burdens of each decision. Some healthcare professionals and bioethicists argue that considering the child’s “best interests” in isolation is unrealistic given that parents also have to balance the needs and interests of other family members. The lives of physicians, health care professionals and other stakeholders are not affected by these decisions in the same way.
as the lives of Ashley’s family members. Therefore the parents’ decisions, while not without limits, must carry significant weight.

**Ethical Issues for Stakeholders: Physicians and Healthcare Professionals**

People with disabilities are high users of medical services, and most physicians are not trained to deal with the complex issues that accompany disability. Patients with permanent disabilities have historically posed particular challenges for health care professionals. People with disabilities often require health interventions to prevent diseases and complications from occurring, but invariably have environmental and social components that complicate their degree of function, social participation, and quality of life. Many physicians have never heard of the social model of medicine, or George Engel’s biopsychosocial model. Nor are they familiar with disability resources and specialists (such as physiatrists, independent living centers, etc.). Finally, numerous studies have demonstrated that health care professionals are not very accurate at predicting what life with disability is like and, indeed, consistently underestimate the quality of life as expressed by people who live with disabilities and chronic conditions.

Physicians have responsibility to frame treatment options based on principles of evidence-based medicine (EBM). EBM involves “the conscientious, explicit, and judicious use of current best evidence in making decisions about the care of individual patients. The practice of evidence-based medicine means integrating individual clinical expertise with the best available external clinical evidence from systematic research.”

It is also interesting to consider the assumptions of benefit made in light of more recent data on growth curves for children with moderate to severe cerebral palsy (CP), a diagnosis that is more akin to Ashley’s condition than the study group of nondisabled, tall adolescent girls. The researchers found that these children with CP, though often smaller than their nondisabled peers, tended to have better health and social participation if they had better growth. Clearly much is not known about the effects of growth on health and social participation for children with significant developmental disabilities.

Though in general, medical professionals do not offer treatments where the risks exceed the expected or known benefits, exceptions do occur. An example might be when a disease process, such as cancer, may result in death without treatment. There may even be times when some risky treatments—such as mastectomy—are initiated prophylactically, because the risk of developing breast cancer is believed to be very high (such as for a woman who carries the BRCA genes). The decision to perform surgery prophylactically vs. increasing the breast cancer screening protocols is not universally accepted, though, and the fact that a woman (i.e., the decision is not offered to children) can weigh the risk and benefits and make her own decision based on her values is a critical variable in the decision making.

There are also examples of prophylactic interventions that are widely adopted on behalf of children—like immunization practices. These are practices in which the benefits are believed to be very high (though the actual risk of infectivity may be low) and the risk from the treatment is also believed to be very low. Ashley does not easily fit into either category. One could argue that she does not have a serious disease that warrants risky medical interventions, and as a prophylactic intervention, the risk of the Ashley Treatment is substantial and not proportionate to the identified problem. Others may believe, as Ashley’s physician’s concluded, that “these risks do not seem to be unreasonable and are not out of line with the risks of other medical interventions these children undergo.”

The final risk/benefit analysis is contingent upon how the problem is defined, how the risk is assessed and how alternative options are identified.

**Ethical Issues for Stakeholders: Bioethics Committees and/or Consultants**

Ethics committees and consultation services were established as advisory bodies to provide a forum for discussion of complex issues in clinical care. Since Karen Ann Quinlan, the courts have asked that hospital-based ethics committees be formed and serve as the primary resource to families and health care providers for ethical dilemmas, rather than the courts. “[This would allow] some much needed dialogue regarding these issues and [force] the point of exploring all of the options
Disability organizations have raised concerns that the ethics committee process in the Ashley case prevented the sort of due process protections that the judicial system would have provided. Not Dead Yet, a disability activist organization opposed to assisted suicide and euthanasia, issued a statement condemning the ethics committee and viewed the Ashley Treatment as doing an “end-run” around Ashley’s constitutionally guaranteed right of due process.28 The Disability Rights Education and Defense Fund believes Ashley was denied her “basic human rights through draconian interventions to her person” and the conduct of the physicians and ethics committee should be “widely questioned.”29 Three other groups who have long-served children with disabilities issued statements. The American Association on Intellectual and Developmental Disabilities categorizes the Ashley Treatment as “unjustifiable nontherapy” and asks why an independent legal advocate or expert in disability rights was not part of the review process.30 United Cerebral Palsy and The Arc, in their joint statement, believed the counsel on behalf of the ethics committee to the parents and physicians was “insufficient . . . since they failed to account for Ashley's fundamental rights as a person with intellectual and developmental disabilities . . . consideration of Ashley and her stake in her own civil and human rights was absent.”31 The Disability Rights Washington (formerly the Washington Protection and Advocacy System) criticized the Ethics Committee process further, agreeing that a court order was legally required before the hysterectomy could be performed.32 As part of the settlement agreement with Seattle Children’s Hospital they recommended that a disability rights advocate be included in future deliberations regarding the Ashley Treatment or growth limiting interventions for children with developmental disabilities.33

Stakeholder representation is critical but this recommendation is not without complexity. Somebody who has a lived experience of disability may be able to authentically represent a different perspective than the professionals who typically sit on hospital ethics committees. However, not everybody with a disability thinks alike or holds the same opinion. How should the disability representative be selected? And can a cognitively intact adult with a disability represent Ashley’s interests any better than her parents or others on the committee? Certainly, every hospital ethics committee must think critically about these questions, the issue of representation, and the group process.

Whereas some have critiqued the ethics committee proceedings, others within the bioethics community have supported the process that took place in Washington and lauded Diekema and Gunther for publishing this case. Three bioethicists writing in Scientific American.Com (Joel E. Frader, MD, Norman Post, MD, MPH, and Benjamin Wilfond, MD) “came down firmly on the side of the parents and the decision of the original ethics board” with a few reservations.32 Norm Frost writes, “whatever disagreements critics may have, it is not possible to say this decision was made casually or quickly, without careful consideration for the relevant facts and arguments.”32 They believe that the child’s best interests remained the focus of the ethics committee’s decision and that going to court for permission on every invasive procedure involving children would be untenable.

Ethical Issues for All Stakeholders

A critical question in the Ashley case is whether it is ever justified to surgically alter the body of an otherwise healthy and nonconsenting child? Quality-of-life criteria were the foundation for the arguments put forward by the parents and physicians.1,2 Although there are examples of quality-of-life criteria justifying surgical interventions, such as in esthetic surgery, an important criteria is who defines the “quality-of-life” goal of treatment. People who elect to have esthetic surgery make the decision for themselves based on their assessment of quality-of-life benefits and medical risks. Quality-of-life criteria have also been used in decisions to treat children, such as the use of synthetic growth hormone for boys who are constitutionally short (but not growth hormone deficient), or high-dose estrogen for girls who were constitutionally tall (ironically the only limited experience with this treatment as applied in the Ashley case), as short boys and tall girls have been considered socially and culturally undesirable.33,34 The use of medical treatments for such nonmedical, quality-of-life indications have been considered controversial, and in general, have fallen out of favor.

Medical historian, Alice Dreger,35 (p261) who has studied conjoined twins and people with intersex conditions offers the following advice to parents contemplating such surgeries:
Make sure you ask yourself and your surgeon more than once: Given the risk, should you ever choose, on behalf of someone else (even your own child), a surgery offered purely for psychological and social reasons? Surgeons often tell me ‘you can’t change society so you have to change children’s bodies,’ but clearly in many cases we do try to change society. We’ve tried to eliminate sexism that tries to keep girls down just because their bodies are different than boys, we’ve tried to eliminate racism that keeps some kids down just because their bodies are darker than others. So consider the possibility that, instead of changing your child’s body, we should start expecting others to change their minds.

The use of quality-of-life criteria when making decisions on behalf of people who cannot speak for themselves is further problematic because of the limited ability of third parties to predict what constitutes a “good” quality of life for others. The social model of disability would question whether the identified problem in this case is intrinsic to Ashley’s body or embedded in a lack of environmental supports, negative social attitudes and the way others see her or frame her possibilities. Evidence for this concern is implicit—both in the parents’ blog and public commentaries, one of which states, “given Ashley’s mental age a nine and a half year old body is more appropriate and more dignified than a fully grown female body.” In apparent support of this position Ashley’s parents quote George Dvorsky, a member of the board of directors for the Institute for Ethics and Emerging Technologies, “The estrogen treatment is not what is grotesque here. Rather, it is the prospect of having a full-grown and fertile woman endowed with the mind of a baby.”

What if the fundamental issue driving the Ashley Treatment is this sentiment—the fact that society is uncomfortable with adults who have cognitive disabilities? If so, should health care professionals ever sanction or be complicit in using medical treatments to alter the bodies of people with disabilities to make them more acceptable, more lovable, or more comfortable to others? And if not, should there be prohibitions on certain types of medical treatments for children and those who cannot consent for themselves?

SUMMARY
Ashley’s treatment undoubtedly encompasses issues related to her biology, her psychological well-being, her cultural/familial environment and sociopolitical factors. Her impairments, the restricted options available to her family, the limits of medicine and the history of discrimination against children with cognitive/developmental delays all come together to make this case particularly poignant and compelling. Given our unique training and experience, it is imperative that physiatrists, as well as other specialists in disability, address these concerns and, along with other stakeholders, be intimately involved in this ethics debate.

ACKNOWLEDGMENTS
The authors would like to thank Debjani Mukherjee PhD, Carmen Cicchetti MA, Elliot J. Roth MD, and Raymond H. Curry MD for their critical review of the manuscript.

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CME Self-Assessment Exam

Answers

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CME Article Number 2:
J. A. Saveika, et al.

1. B
2. A
3. A
4. A
5. D

Dec 2007

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Exacerbation of Habitual Dislocation of Ulnar Nerve by Concurrent Dislocation of Triceps Muscle: Complementary Role of Dynamic Ultrasonography to Electrodiagnosis

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A 50-yr-old woman with past medical history of left elbow fracture in childhood presented with 3 mos of numbness and tingling of the left ring and little fingers. Physical examination was remarkable for cubitus varus deformity, atrophy and weakness of the left hand intrinsic muscles, and a positive Tinel sign over the left retrocondylar groove. During elbow flexion, snapping of a cordlike structure was noted bilaterally over the medial epicondyle.

Electrodiagnosis demonstrated markedly slow conduction velocity of the left ulnar nerve across the retrocondylar groove. Fibrillation potentials and polyphasic motor unit action potentials were noted in the left first dorsal interosseous, abductor digiti quinti, flexor digitorum profundus, and flexor carpi ulnaris muscles. These findings were compatible with entrapment neuropathy of left ulnar nerve at the retrocondylar groove.

To evaluate the “snapping” structure over the medial epicondyle, dynamic ultrasonography was performed using a broadband linear transducer with medium frequency of 10 MHz (LogiqE, General Electric, Inc). The left ulnar nerve was markedly enlarged and hypoechoic compared with the contralateral side (Fig. 1). At 70 degrees of elbow flexion, dislocation of the left ulnar nerve over the medial epicodyl and sonographic imaging revealed concomitant dislocation of the triceps muscle pushing the ulnar nerve and producing an appreciable snap (Fig. 2). Ultrasonography of the contralateral elbow revealed habitual dislocation of the ulnar nerve, without clinical or electrodiagnostic findings of nerve dysfunction. On the basis of the electrodiagnostic and ultrasonographic findings, snapping triceps syndrome with resulting ulnar nerve entrapment at the elbow was diagnosed. The patient was referred for operative management, including transposition of the ulnar nerve and medial triceps, as well as corrective valgus osteotomy of the distal humerus.

A transient snapping sensation during elbow flexion can be caused by either dislocation of the ulnar nerve or the medial portion of the triceps muscle.\(^1\) In this case, a palpable snap over the medial epicodyl was initially interpreted as a habitual dislocation of the ulnar nerve. Dynamic ultrasonography differentiated concurrent dislocation of the triceps muscle as well as the ulnar nerve on the symptomatic side from isolated dislocation of the ulnar nerve on the asymptomatic side.\(^2\) This case illustrates the complementary role of ultrasonography to electrodiagnosis in evaluating underlying anatomic structures of entrapment neuropathy such as snapping triceps syndrome, posterior interosseous nerve syndrome, or tarsal tunnel syndrome.

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All correspondence and requests for reprints should be addressed to Mooyeon Oh-Park, MD, Department of Rehabilitation Medicine, Albert Einstein College of Medicine, Montefiore Medical Center, 111 E 210th Street, Bronx, NY 10467.
Re: Introduction to Nanotechnology: Potential Applications in Physical Medicine and Rehabilitation

To the Editor: We find the article titled “Introduction to nanotechnology: potential applications in physical medicine and rehabilitation” by Gordon and colleagues (March 2007) interesting and timely.1 Scientist now attempt to manipulate matter at the scale of atoms and molecules, using the term nanotechnology to revolutionize manufacturing across all industry sectors, including tissue engineering, drug delivery, imaging, diagnostics, surface texturing, and biointerfaces that could impact the practice of physical medicine and rehabilitation in the future. Governments and conglomerates must establish strict codes of ethics for nanotech products to avoid future mismanagement of this novel technology. Lack of appropriate ethical codes of conduct in this science may lead to disasters to mankind as well as nature. Perhaps it is time to remember that natural organisms harbor excellent nanotech protection shaped by millions of years of natural selection—lotus leaves possess micro- and nanotextures on the surface. Similarly, animals from bees to birds have unique nanotexturing that engenders water repellency and lack of adhesion. Prehistoric cave painters apparently used nature’s nano materials in their painting processes.

Globally, industry and governments spend nearly $10 billion on nanotech research and development annually (Table 1). The National Science Foundation in the United States predicts that nanoscale technology will capture a $1 trillion market by 2011. Thousands of tons of nanomaterials are already being produced each year, and more than 300 claimed nanotechnology products have surfaced on the market.2 Nano particles like nano paint come when the general public has great concern regarding genetically modified organisms and toxic chemical residues in human and pet foods. Thus, the public in developed and developing nations may undoubtedly demand from their governments more transparency and safer nanotechnology.3 Unfortunately, there are no ethical and professional regulations for large- or small-scale nanotech industries, and no impact studies have been mandated to assess exposure to engineered nanomaterials in air, water, and terrestrial ecosystems. Because nanotechnologies are diverse and exposures to nonmaterial will vary widely, assessing exposure and potential impacts on health or the environment will require a multidisciplinary approach for assessments.

Among the large array of nanomaterials and processes, the quantum dots are of particular importance for their possible therapeutic and diagnostic medical applications. These nanoparticles consist of a metalloid core and a shell that surrounds the core and renders the quantum dots’ bioavailability. The further addition of a specific coating can give the quantum dots a desired bioactivity, which is essential for promising medical applications such as targeted drug delivery and in vivo biomedical imaging. Human exposure to quantum dots may result from environmental, workplace, and therapeutic exposure. There may be a risk of bioaccumulation of these materials within organs and tissues (e.g., in lungs) with still unexplored or underexplored health risks. Introduction of quantum dots into the environment may occur via waste streams from industry, research, and clinical settings. The persistence of these materials in the environment may be quite long, and the environmental exposure will depend on the partition of quantum dots between water, air, and various soil types.4 Particular impacts of nanomaterials are foreseen on the ecology of soils, which often already contain and move around small particles. Nanoparticles could easily be absorbed by earthworms, possibly allowing them to move up the food chain and reach humans.5

Global understanding of nanotechnology-specific risks is essential if large and small industries are to operate on a level playing field and if developing economies are not to be denied essential information on designing safe nanotechnologies. The ethical issues fall into the areas of equity, privacy, security, environment, and metaphysical questions concerning human–machine interactions. A wide debate is needed on the implications and democratic control of nanotechnologies worldwide, to restrict nanotechnology before it becomes a lawless science. The debate should not be restricted to meetings of expert scientists or targeted on human/environmental health and safety issues. Ethical implications of nanotechnology must be addressed by people from all walks of life.

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Re: Sacroiliac Joint Pain: Anatomy, Biomechanics, Diagnosis, and Treatment

To the Editor: Foley and Buschbacher2 are to be commended on their informative as well as thorough review of the sacroiliac joint (SIJ) as a pain generator (December 2006). As they readily acknowledge, “the literature on SIJ pain is complex and controversial.” Three decades ago, when we evaluated the SIJ as a source of lumbosacral and pelvic pain, it was also underappreciated, as it remains so even today.2

Although Dreyfuss et al.3 state in their review “that no physical examination test [has] demonstrated diagnostic value and that SIJ pain is resistant to identification by historical and physical examination,” our experience in this regard would suggest otherwise. Although we would agree there is no one clinical or laboratory test that is pathomonic of SIJ dysfunction, an appropriate history and physical examination buttressed by dynamic pelvic x-rays and an immediate reduction in pain in response to an anesthetic injection of the joint can be diagnostic. In the supine position, an asymmetric restriction of hip external rotation on the symptomatic side associated with increased pain over the SIJ as it is passively rotated into abduction is also a frequent clinical finding (Fig. 1). A standing pelvic x-ray followed by alternate-leg weight bearing may also demonstrate excessive instability of the pubic symphysis, often exceeding 3 mm (Fig. 2). Usually, this slip is greater on the side of the symptomatic SIJ. Symphyseal instability mirrors similar rotary luxation at the symptomatic SIJ as this articulation responds to shifts in the center gravity horizontally as well as vertically during normal ambulation (Fig. 3).

An immediate reduction in SIJ pain after a combined injection of a steroid and Xylocaine is also diagnostic.

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FIGURE 1 Asymmetric restriction of hip external rotation on the symptomatic side is associated with increased pain over the sacroiliac joint as it is passively rotated into abduction.
Except in the presence of extreme obesity, this procedure can easily be accomplished without x-ray guidance. Entrance to the articulation itself can be assured by noting restricted side-by-side movement of the syringe as limited by the congruent joint surfaces. As a caveat, it is important to note that SIJ may not be the primary pain generator. Instead, it may be a referred site of discomfort, both via its direct innervation and secondary to more remote mechanical problems—among others, a degenerative disc at L5–S1 and/or spinal stenosis, as well as degenerative disease of the hip.

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Response Letter

The Authors Respond: The authors appreciate Dr. LaBan’s thoughtful comments. We agree that although no single physical exam test is particularly predictive, a panel of three or more tests can be extremely helpful. The tests described by Dr. LaBan may prove useful to those examining patients with sacroiliac joint pain. Hopefully, further research will be performed to test those hypotheses. They make sense to the authors, but they have not yet been validated in the literature.

Dr. LaBan’s caveat regarding other regional pain generators that masquerade as SIJ pain is a good one that should be heeded by all practicing clinicians.

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Re: The Myofascial Trigger Point Region: Correlation Between the Degree of Irritability and the Prevalence of Endplate Noise

To the Editor: The article “The myofascial trigger point region: correlation between the degree of irritability and the prevalence of endplate noise” by Kuan et al.1 (March 2007) is a valuable addition to the current body of knowledge surrounding myofascial pain. The authors suggest a possible mechanism of action proposed originally by Simons et al.2 in which hypoxia and ischemia are related to local release of inflammatory substances, which may sensitize muscle nociceptors. We would like to add that our recent study3 in 2005 also supports this contention.

We used a novel microdialysis technique to sample muscle with and without myofascial trigger points for levels of protons, inflammatory mediators, catecholamines, neuropeptides, and cytokines. The results show distinct local biochemical differences between subjects with a spontaneously painful myofascial trigger point (active MTrP) in the upper trapezius muscle compared with those with a nonpainful trigger point (latent MTrP) and those without a trigger point. Analysis for pH demonstrated that active MTrPs possessed relatively higher acidic levels (lower pH) than the other muscle groups, which could be associated with conditions attributed to hypoxia and ischemia. Similarly, levels of bradykinin, serotonin, norepinephrine, substance P, CGRP, TNF-α, and IL-1β were all at relatively higher concentrations in active MTrPs.

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FIGURE 2
Standing pelvic x-ray showing excessive instability of the pubic symphysis.

FIGURE 3
Diagram showing rotary luxation at the symptomatic sacroiliac joint during normal ambulation.
We feel that the combination of these two studies provides good support to the proposed mechanism of myofascial trigger point pain.

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Response Letter

The Authors Respond: We highly appreciate the comments by Shah et al. We also regret that we have not cited their paper1 as one of the references in our article.2 Both their work1 and ours2 have definitely contributed substantially to increasing the credibility of myofascial trigger point (MTrP). The former has further confirmed the energy crisis hypothesis,3 and the latter has proven the correlation between MTrP and endplate noise.4,5 The great pioneer in MTrP, David G. Simons, developed these two important theories more than 10 yrs ago.

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DOI: 10.1097/01.phm.0000297445.62886.08

Re: Characteristics and Treatment of Headache After Traumatic Brain Injury

To the Editor: The authors of “Characteristics and Treatment of Headache After Traumatic Brain Injury” (Lew et al.,1 July 2006) are to be complimented on their comprehensive review of “posttraumatic headache (PTH).” They readily acknowledge the present confusion inherent in both the “accepted terminology” and the exact pathogenesis of “cervicogenic headache” pathology when both intracranial and cervical spine trauma have occurred simultaneously.

Although controversy in this regard persists, in our own 40-yr clinical experience, posttraumatic occipital neuralgia (PTON) is all too often the principle causal agent in cases of persistent PTH. The discomfort of PTON is located primarily in the occipital area, often radiating to the vertex as a direct consequence of injury to the greater and/or lesser occipital nerves at their C1–C2 spinal roots of origin.

When digital pressure is applied to these nerves over the occipital notch, the patient may experience either hyper- and/or hypoesthesia in the occipital nerve’s distribution. Thereafter, an occipital nerve block can be both diagnostic and therapeutic in confirming the origin of the headache by alleviating the symptoms of the PTON.

A 25-gauge needle in this procedure is inserted 3.5 cm inferolaterally to the occipital protuberance, and a combination of both bupivacaine hypochloride (Marcaine) and a corticosteroid (1 ml of each) is injected. In a review of ten PTH patients, Lew report a good response to a greater occipital nerve block in eight patients and, in an additional two patients, a partial response.2

Additional symptoms often accompanying PTON include, among others, a “light and sound hypersensitivity,” as well as vertigo, nausea, and facial hyperesthesia. At the upper spinal cord levels, the fibers of the descending tract of CN V, accompanied by the tracts of CN VII, IX, and X, interdigitate with the ganglion cells of CI and CII. Subsequently, these tracts cross the midline together and progress retrogradely as the ascending tract of CN V. At the C1–C2 spinal levels, this tract is vulnerable to trauma, with the potential of inciting brainstem symptoms.

As the authors themselves conclude, “headaches are multifactorial and involve a combination of central and peripheral mechanisms.” We would agree that a definitive diagnosis as related to intracranial and/or extracranial pathology remains paramount to efficient and appropriate treatment.

Myron M. LaBan, MD, MMSc
Royal Oak, Michigan
Response Letter

The Authors Respond: The authors appreciate the insightful comments by Dr. Myron M. LaBan regarding our recent article on headache after traumatic brain injury (TBI).1 Incidentally, after publication of this manuscript, we have received feedback from private practitioners of different specialties, as well as academicians, who have echoed our perception that most headaches involve a combination of central and peripheral mechanisms. They also share our observations regarding the heterogeneity in terminology and the paucity of well-controlled studies in this field. The consensus is that clinicians should perform a comprehensive and systematic evaluation before initiating appropriate treatment. What we propose in Figure 2 is one possible algorithm that can be expanded on, according to the local resources and level of expertise of the individual clinician. As described by Dr. LaBan, occipital nerve block is one example of a dual diagnostic and therapeutic approach that is relatively time-efficient to perform and that could prove to be useful to patients. Like all other methods we have alluded to, this procedure deserves a focused and systematic evaluation of elder asymptomatic patients (60 yrs of age and above) showed an incidence of 28% full-thickness tears.3

Incidentally, after publication of this manuscript, we have received feedback from private practitioners of different specialties, as well as academicians, who have echoed our perception that most headaches involve a combination of central and peripheral mechanisms. They also share our observations regarding the heterogeneity in terminology and the paucity of well-controlled studies in this field. The consensus is that clinicians should perform a comprehensive and systematic evaluation before initiating appropriate treatment. What we propose in Figure 2 is one possible algorithm that can be expanded on, according to the local resources and level of expertise of the individual clinician. As described by Dr. LaBan, occipital nerve block is one example of a dual diagnostic and therapeutic approach that is relatively time-efficient to perform and that could prove to be useful to patients. Like all other methods we have alluded to, this procedure deserves a focused and systematic evaluation of elder asymptomatic patients (60 yrs of age and above) showed an incidence of 28% full-thickness tears.3

In addition, Neer5 and Hawkins–Kennedy6 impingement tests are not enough for diagnosis of SIS. These tests may be positive in other shoulder disorders. SIT should be performed as a reference test for differential diagnosis of SIS.5,7–9 Neer5,7 identifies SIT in his reports as a most useful method of separating impingement lesions from other causes of shoulder pain. SIT does not investigate the anatomopathologic condition in RC, but it depresses the pain generators in the subacromial area where the impingement occurs. Obvious relief of pain and almost total improvement in passive and/or active shoulder range of motion after SIT suggest that the test is positive and discriminates SIS from other shoulder pathologies.

As a result, MRI is not the gold standard for diagnosis of SIS. SIS is a clinical diagnosis that is done with SIT and impingement tests.

REFERENCES


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Re: Shoulder Impingement Syndrome: Relationships Between Clinical, Functional, and Radiologic Findings

To the Editor: We read with interest the article by Ardic and colleagues.1 The authors detected the relationship between clinical, functional, and radiologic diagnostic (ultrasonography, magnetic resonance imaging [MRI]) measures in patients with subacromial impingement syndrome (SIS) and compared the diagnostic performances of ultrasonography and MRI. In this study, MRI has been used as a gold standard. However, MRI mainly evaluates the anatomopathologic conditions in the rotator cuff (RC) and surrounding structures. It has also been shown that a significant number of asymptomatic shoulders may reveal abnormal internal signals and even complete ruptures of RC tendons, by MRI.2,3 These tears were increasingly frequent with advancing age and were compatible with normal subjects. MRI evaluation of elder asymptomatic patients (60 yrs of age and above) showed an incidence of 28% full-thickness tears.3

Frost et al.4 report that age was related to MRI based on supraspinatus pathology more so than clinical signs of impingement.

In addition, Neer5 and Hawkins–Kennedy6 impingement tests are not enough for diagnosis of SIS. These tests may be positive in other shoulder disorders. SIT should be performed as a reference test for differential diagnosis of SIS.5,7–9 Neer5,7 identifies SIT in his reports as a most useful method of separating impingement lesions from other causes of shoulder pain. SIT does not investigate the anatomopathologic condition in RC, but it depresses the pain generators in the subacromial area where the impingement occurs. Obvious relief of pain and almost total improvement in passive and/or active shoulder range of motion after SIT suggest that the test is positive and discriminates SIS from other shoulder pathologies.

As a result, MRI is not the gold standard for diagnosis of SIS. SIS is a clinical diagnosis that is done with SIT and impingement tests.

REFERENCES


2. Chandnani V, Ho C, Gerharter J, et al: MR findings in asymptomatic...
Response Letter

The Authors Respond: Many thanks for your interest to our article. First of all, our studies were done by an upper-extremity rehabilitation team consisting of experts in this area using standardized evaluation and therapy systems/algorithms.

We have to use comprehensive diagnostic and therapeutic tools for patients’ referral to shoulder rehab, as we mentioned before. Special shoulder tests commonly performed by upper-extremity professionals can be divided for instability, impingement, labral lesions, scapular stability, muscle tendon pathology, brachial plexus, and thoracic outlet syndrome. Common shoulder impingement tests are the Neer–Walsh, Hawkins–Kennedy, and posterior internal impingement tests. We used the first and the second impingement tests because the third is found primarily in overhead athletes and workers.

The positive Neer–Walsh impingement test suggests rotator cuff (RC) pathology like the positive Hawkins–Kennedy impingement test. If one of these tests elicits pain, subacromial impingement test (SIT) with 10 ml of 0.1% lidocaine to the subacromial space will eliminate the pain of repeat testing. Because of medicolegal issues and the approval procedure of this study by the IRB, we didn’t choose the subacromial impingement test to assist in the diagnosis. It is not clear that these injections have proven useful in directing care toward successful outcomes, possibly because of the confounding placebo factor. Actually, Henkens et al. report that injections in subacromial bursa were inaccurate, with false-positive and -negative results in their randomized controlled MRI study. We didn’t prefer SIT, because it is invasive; it requires patient’s approval and is only 66% accurate. Our opinion for the diagnosis of subacromial impingement syndrome (SIS) was noninvasive clinical and radiologic work-ups.

We selected patients that had no history of trauma and had shoulder pain of >3 mos, were unresponsive to analgesic medication after 3 wks, and had a diagnosis of suspected SIS waiting for physical therapy and shoulder rehab. It is ridiculous to make clinical and radiologic assessments for asymptomatic shoulders. We carefully evaluate the patients with chronic shoulder pain demographically, clinically, and radiologically for the treatment options. Diagnosis and treatment should be based on combination of these data.

As we state in our article, “MRI has been used as the gold standard because our patients did not want arthroscopic evaluation.” In addition, MRI has been accepted as the “best imaging tool” in many shoulder problems, including RC, labrum, capsule, biceps tendon/anchor, osseous structures, arthritis, and neural impingement. It is well known that MRI is the only technique that can accurately image the early changes of this condition, in particular bursal thickening and effusion (subacromial bursitis), edema, and inflammatory changes of the rotator cuff and its tendons. SIS is a clinical diagnosis, and MRI provides us with critical information regarding the size and location of a tear, the specific tendons involved, the degree of muscular atrophy and tendon retraction, and the quality of the torn edges.

Neer’s impingement concept depicts the syndrome as a lifelong process with three stages. Stage I impingement consists of reversible edema and hemorrhage typically in active patients ≤25 yrs. Stage II implies fibrosis and tendinitis. Stage III represents degeneration and rupture often associated with osseous changes most commonly in patients >40 yrs. Therefore, early diagnosis and treatment are critical to prevent progression and improve shoulder function. Frequently, however, clinical signs and symptoms are nonspecific, and the diagnosis is often delayed until a full-thickness defect in the RC has developed. Symptomatic elderly patients may have stage III SIS mainly treated by shoulder rehab. However, asymptomatic individuals are not our interest area.

Finally, there were very limited numbers of randomized controlled trials for SIS using clinical and MRI variables. MR/MR arthrography are still the best imaging tools in many shoulder problems. MRI has proved to be highly sensitive and accurate for the diagnosis of stage II–III SIS, with excellent correlation of arthroscopic and/or miniopen surgical findings. Specified clinics in shoulder rehab should use comprehensive diagnostic and therapeutic work-ups.

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REFERENCES


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In the article by Ersoz et al., published in the September 2007 issue of the *American Journal of Physical Medicine & Rehabilitation*, Tables 1 and 4 should appear as follows:

### TABLE 1. Comparison of the stroke subgroups with respect to demographic and clinic parameters for the frequencies of symptomatic urinary tract infection

<table>
<thead>
<tr>
<th>Factor</th>
<th>Frequency (%)</th>
<th>P</th>
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</thead>
<tbody>
<tr>
<td>Age, yrs</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt;65</td>
<td>11/56 (19.6)</td>
<td>0.067</td>
</tr>
<tr>
<td>≥65</td>
<td>19/54 (35.2)</td>
<td></td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>10/49 (20.4)</td>
<td>0.147</td>
</tr>
<tr>
<td>Female</td>
<td>20/61 (32.8)</td>
<td></td>
</tr>
<tr>
<td>Level of education</td>
<td></td>
<td>0.055</td>
</tr>
<tr>
<td>Ungraduated</td>
<td>18/46 (39.1)</td>
<td></td>
</tr>
<tr>
<td>Primary school</td>
<td>9/44 (20.5)</td>
<td></td>
</tr>
<tr>
<td>Secondary/high school or university</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Type of lesion</td>
<td></td>
<td>0.858</td>
</tr>
<tr>
<td>Hemorrhagic</td>
<td>8/28 (28.6)</td>
<td></td>
</tr>
<tr>
<td>Ischemic</td>
<td>22/82 (26.8)</td>
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</tr>
<tr>
<td>Side of lesion</td>
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<td>0.354</td>
</tr>
<tr>
<td>Right</td>
<td>6/29 (20.7)</td>
<td></td>
</tr>
<tr>
<td>Left</td>
<td>24/81 (29.6)</td>
<td></td>
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<tr>
<td>Bladder emptying method</td>
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<td>0.041</td>
</tr>
<tr>
<td>Spontaneous</td>
<td>23/96 (24.0)</td>
<td></td>
</tr>
<tr>
<td>Indwelling catheter</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Residual urine, &gt;50 ml</td>
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<td>0.039</td>
</tr>
<tr>
<td>Absent</td>
<td>15/77 (19.5)</td>
<td></td>
</tr>
<tr>
<td>Present</td>
<td>8/19 (41.2)</td>
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<tr>
<td>Functional ambulation class</td>
<td></td>
<td>0.147</td>
</tr>
<tr>
<td>≤2</td>
<td>20/61 (32.8)</td>
<td></td>
</tr>
<tr>
<td>&gt;2</td>
<td>10/49 (20.4)</td>
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<tr>
<td>Brunnstrom stage class of upper extremity</td>
<td></td>
<td>0.012</td>
</tr>
<tr>
<td>1—2</td>
<td>23/59 (39.0)</td>
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</tr>
<tr>
<td>3—4</td>
<td>3/26 (11.5)</td>
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</tr>
<tr>
<td>5—6</td>
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<td>Brunnstrom stage class of lower extremity</td>
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<tr>
<td>3—4</td>
<td>9/44 (20.5)</td>
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</tr>
<tr>
<td>5—6</td>
<td>5/27 (18.5)</td>
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</tr>
</tbody>
</table>

### TABLE 4. Microorganisms isolated in urine (≥10⁵ CFU/ml) and their frequencies in all stroke patients, in patients who are voiding spontaneously, and in patients with indwelling catheters

<table>
<thead>
<tr>
<th>Microorganism</th>
<th>Spontaneous (%) (n = 32)</th>
<th>Indwelling Catheter (%) (n = 11)</th>
<th>All Patients (%) (n = 43)</th>
</tr>
</thead>
<tbody>
<tr>
<td><em>Escherichia coli</em></td>
<td>19 (59.4)</td>
<td>6 (54.5)</td>
<td>25 (58.1)</td>
</tr>
<tr>
<td><em>Klebsiella pneumoniae</em></td>
<td>7 (21.9)</td>
<td>0 (0)</td>
<td>7 (16.3)</td>
</tr>
<tr>
<td><em>Pseudomonas aeruginosa</em></td>
<td>0 (0)</td>
<td>4 (36.4)</td>
<td>4 (9.3)</td>
</tr>
<tr>
<td><em>Staphylococcus aureus</em></td>
<td>3 (9.4)</td>
<td>1 (9.1)</td>
<td>4 (9.3)</td>
</tr>
<tr>
<td><em>Proteus mirabilis</em></td>
<td>3 (9.4)</td>
<td>0 (0)</td>
<td>3 (7.0)</td>
</tr>
</tbody>
</table>

REFERENCE