Friedreich’s Ataxia and Gait Changes through Participation in Therapeutic Horseback Riding

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ABSTRACT
This study examined potential benefits from participation in therapeutic horseback riding for an individual with Friedreich’s Ataxia (FA). FA is an autosomal recessive neurological disorder with a side effect being gait ataxia. The participant’s gait was analyzed using Dartfish™ video software before and after two separate 6-week riding sessions, separated by a five-month lay-off. A significant difference in stride length ($p < 0.05$) was found, with a decrease occurring in the second riding session (0.99 ± 0.10 m to 0.74 ± 0.19 m). Analysis of the other variables revealed no significant differences in gait before and after each individual riding session, nor were they observed from the first day to the last day of each six week session, or intersession observations. The decrease in stride length may indicate a decrement in stability as the disease progresses, but the lack of significant differences in joint angles may indicate therapeutic horseback riding helped prevent some of the decrements caused by FA.

Key Words: gait analysis, equine assisted therapy, neurological disorder

INTRODUCTION
Friedreich’s Ataxia (FA) is a rare, autosomal recessive hereditary disorder (13) caused by a defect in a recessive gene labeled FXN, affecting approximately 1 in every 50,000 people in the United States (6). The specific clinical and pathological manifestations of FA are said to be attributed to the degeneration of the posterior columns of the spinal cord. Although FA is not common, it is the most common form of inherited ataxia. Gait disorders are the dominant symptom of FA (7), and the average age of expression is generally traced back to the age of 15 even though the average age for medical diagnosis is usually later in life. The gait disorders continue to progress until it is necessary for the individual to rely on a wheelchair for mobility, which occurs approximately 15 years after the onset of symptoms (approximately by the age of 30). Neuropathology of FA is demonstrated by progressive ataxia, loss of coordination and reflexes, muscle weakness, dysarthria, extensor plantar responses, and is often associated with scoliosis and foot deformity (13).

Individuals with FA are at greater risk for other medical problems such as diabetes or heart conditions (13), but those conditions are not generally the primary concern. The primary concern for individuals with FA is motor processing responsible for mobility, and physical therapy is the only suggested treatment to prolong independent mobility and maintain optimal functioning of the limbs for as long as possible. Even though it is generally understood that gait training has been the only successful method used to delay the need for a wheelchair (4), there are limited measures used to assess changes in gait for individuals with FA.

Additionally, there are limited methods used to measure the overall progression of FA, and the majority of those available are derived from assessment protocol used for individuals with Multiple Sclerosis (MS). One method of quantifying the progression of FA has been done in a clinical setting through the use of the Friedreich’s Ataxia Rating Scale (FARS), which has five separate subscales based on a clinical-examination. Although the FARS was designed specifically for diagnosing individuals with FA, it was designed for a clinical setting and is not practical for use in a functional setting and may not be sensitive enough to measure progression or changes in ambulation (6). A more functional mobility based assessment has been used to assess the progression of MS using performance measures derived from the Multiple Sclerosis Functional Composite (MSFC), which is a battery of measures. There are four measures in the MSFC designed to assess: 1) arm movement from a 9-hole peg test, 2) ambulation from a timed 25-foot walk, 3) a speech test because of the potential to develop dysarthria, and 4) a vision test because of the possibility of visual atrophy.

The four measures of the MSFC can be used formatively to assess the progression of FA, but may not be the best assessment to use. Although MS and FA are both classified as degenerative neurological conditions, the etiology and progression are different. Consequently assessment protocol used for one condition may not be appropriate for the other and may lead to inappropriate decisions concerning therapy. There is only one component of the MSFC

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that provides any information regarding ambulation, and the primary concern of the individual with FA is ambulation. There is a need for more measurements to be used for individuals with FA to explain the disorder in greater detail. Providing a more detailed description of the neurological disorder and what an individual with FA is experiencing will assist in the use of more therapeutic methods and alternative therapies that can be used to slow the progression of the condition and allow the individual with FA to maintain independence for a longer period of time.

There is common agreement that the primary clinical presentation of FA is gait disorder or ataxia (7). Gait ataxia is usually the earliest symptom, which is caused by nerve degeneration and loss of proprioception (4). Muscle weakness of the hip extensors and abductors are common and distal limb muscle weakness progresses with the advancement of FA. Thus, slowing the progression of ataxia by maintaining proprioceptive awareness and muscle strength would indicate a delay in the progression of the symptoms of FA. Improvements in proprioceptive awareness has been demonstrated for individuals involved in therapeutic horseback riding, as well as the development of muscle strength in hip extensors and abductors (12).

The North American Riding for the Handicapped Association (NARHA) has defined therapeutic horseback riding as the use of equine-oriented activities to achieve therapeutic goals (12). There are different approaches for using therapeutic horseback riding that are based on the goals and type of disability of the participant. Even though therapeutic benefits are goals of therapeutic horseback riding, the focus is on teaching horsemanship skills. This is in contrast to hippotherapy, which refers to the equine-assisted movement under the direction of a physical or occupational therapist (2). The equine-assisted benefits have been described to occur because the gait of the horse replicates the normal human gait for the rider (3). The specific part of the equine movement that replicates normal human gait includes hip and pelvic rotation, weight shift, and proprioceptive stimulation.

Many other benefits of equine-assisted therapy have been reported, but the vast majority has been in non-juried publications and self-reports. The few studies that have been reported in peer-reviewed journals using an experimental design have been conducted with individuals identified with cerebral palsy (1, 3, 9, 10, 15). Consequently, considerations for using equine-assisted therapy for individuals with cerebral palsy are clearly identified for therapeutic horseback riding instructors, but procedures and considerations for individuals with other conditions are not as clear. The NARHA instructor guide describes specific considerations for individuals with Muscular Dystrophy (MD) and MS, but does not mention Friedreich’s Ataxia. This could lead to therapeutic horseback riding instructors to follow the guidelines and considerations for more common neurologically degenerative conditions, regardless of the actual benefit or effectiveness of the program.

Given that FA is not a common neurological condition, there was only one individual with the diagnosis of FA available to participate in the current therapeutic horseback riding program. Similar measures were conducted on gait for other individuals participating in the same therapeutic horseback riding program with a wide variety of disabilities; such as spinal cord injury, traumatic brain injury, stroke, Down syndrome and cerebral palsy, but the only individual with a degenerative neurological condition was the individual with FA. This study only used the data collected from the individual with FA because the conditions are so dissimilar that any benefit or changes in gait from participation in the therapeutic horseback riding program would not be comparable. The purpose of this case study was to add to the current body of literature to assist in the understanding of the disease progression, and to describe potential benefits for individuals with FA through participation in therapeutic horseback riding. Another purpose of this study was to provide therapeutic horseback riding instructors, or other individuals involved in a variety of movement therapies, guidelines to use when working with individuals with FA.

**CASE STUDY**

**METHOD**

**Participant**

This paper presents a single case study of a 39-year-old male with the clinical diagnosis of FA. He reported having symptoms at age 17, but dismissed his poor balance as clumsiness, and was subsequently diagnosed with FA at the age of 25. Since his diagnosis, he has been involved in a variety of physical therapy, but therapeutic horseback riding has been the only physical therapy he has continued. Consequently, he has been participating in therapeutic horseback riding for the past 7 years and it has been his only form of physical therapy. The participant gave his written consent to participate in this study and to have the results and conclusions published. The protocols were developed to conform to the Human Subjects Review Board of the researchers’ university.

He has used a walker for the past 4 years. Prior to that, he used a cane to help overcome balance and proprioceptive issues. His most recent complaints have been of fatigue, loss of balance, and slowed...
reaction time. He continues to participate in therapeutic horseback riding in an attempt to retain as much independence as possible. Independence and mobility are closely related to quality of life, and the ability to remain active with his family.

Experimental design

The experiment was designed to analyze the participant’s walking gait both before and after each therapeutic horseback riding session, and to analyze any changes in gait that may have occurred over the course of two separate six week riding sessions, approximately five months apart.

After the participant arrived at the horseback-riding center, his walking gait was recorded prior to his therapeutic riding session. With the participant’s left side of the body facing the camera positioned to the side of the participant (sagittal plane), the participant walked 20 meters, turned around, and walked back to the starting position with the right side of his body facing the camera positioned to the side of the participant. This allowed for measurements of joint angles and stride length for both the left and right sides. The participant was instructed to walk at a self-selected walking pace. After recording the first gait session, the participant completed a 30-45 minute therapeutic horseback riding session. After the session was completed, the participant’s gait was recorded a second time.

Biomechanical analysis of gait

Variables. The dependent variables in this study included stride length (m), left and right step length (m), step width (m), and ankle angle, knee angle, and hip angle (degrees) at both heel strike and toe off, measured at two levels for the independent variable of testing time (pre: before riding and post: after riding) and measured at two levels (session one and session two) for the independent variable of testing session.Stride length was defined as the distance, in meters, from left heel strike to left heel strike. Step length was defined as the distance, in meters, from left heel strike to right heel strike (right step length), and from right heel strike to left heel strike (left step length). Step width was defined as the distance, in meters, between the centers of the posterior aspect of the participant’s shoe. Joint angles were measured at each joint axis.

Measurement instruments. To record the participant’s gait, two Sony Handycam (Sony Corporation, Tokyo, Japan) digital camcorders were used. Data was collected at 30 frames per second. One camera was placed to the side of the participant to film his motion in the sagittal plane, and the second camera was placed in front of the participant to film his motion in the frontal plane.

Data processing. To obtain the gait parameters, the video was exported and analyzed using Dartfish video solutions Dartfish video solutions Dartfish Prosuite version 5.5 (Alpharetta, GA) motion analysis software. Two gait cycles were analyzed with the participant’s left leg facing the camera and two gait cycles were analyzed with the participant’s right leg facing the camera in the sagittal plane. These values were then averaged for each trial.

Statistical methods. A 2 x 2 RM-ANOVA (time by session) was conducted to determine if there were significant differences in gait parameters between the different testing times and testing sessions. In the case of significant differences, Tukey’s test of least significant difference (LSD) was conducted to detect post hoc differences. An alpha significance level of 0.05 was set.

RESULTS

Gait Parameters

There was a significant difference in stride length ($F_{1,4} = 10.78, p = 0.03, \eta^2 = 0.729$). Tukey’s LSD revealed a significant decrease in stride length ($p = 0.023$) between the session one pre-test (stride length = 0.95 m) and the session two post test (stride length = 0.704 m), and a significant decrease in stride length ($p = 0.043$) between the session one post-test (stride length = 0.99 m) and the session two post test.

There was no significant interaction between step length (left and right leg) and testing session ($p = 0.817$), or significant difference in step length between the left and right legs ($p = 0.518$). There was a significant difference in step length between testing sessions ($p = 0.029$). Tukey’s LSD revealed significant differences in step length ($p = 0.023$) between the session one pre-test (step length = 0.474 m) and session two post-test (step length = 0.363 m), and significant differences ($p = 0.036$) between the session one post-test (step length = 0.495 m) and session two post test, and significant differences ($p = 0.05$) between the session two pre-test (step length = 0.422 m) and the session two post-test. Step width was only measured for session two. There was no difference in step width between the pre-test and post-test session ($p = 0.074$). The means and standard deviations for stride length, step length, and step width can be found in Table 1.

Joint Angles

There were no significant differences found in any of the joint angles. There were no significant differences between testing sessions and across riding programs for left ankle angle at heel strike ($p = 0.711$), left knee angle at heel strike ($p = 0.458$), left hip angle at heel strike ($p = 0.086$), left ankle angle at
Table 1. Mean (±SD) of the observed distances during the different testing sessions

<table>
<thead>
<tr>
<th>Variable</th>
<th>Session One Pre-Test</th>
<th>Session One Post-Test</th>
<th>Session Two Pre-Test</th>
<th>Session Two Post-Test</th>
</tr>
</thead>
<tbody>
<tr>
<td>Stride Length</td>
<td>0.95 (0.09) m</td>
<td>0.99 (0.10) m</td>
<td>0.84 (0.15) m</td>
<td>0.74 (0.19) m</td>
</tr>
<tr>
<td>Left Step Length</td>
<td>0.49 (0.08) m</td>
<td>0.48 (0.10) m</td>
<td>0.45 (0.08) m</td>
<td>0.37 (0.11) m</td>
</tr>
<tr>
<td>Right Step Length</td>
<td>0.46 (0.04) m</td>
<td>0.51 (0.09) m</td>
<td>0.39 (0.08) m</td>
<td>0.35 (0.10) m</td>
</tr>
<tr>
<td>Step Width</td>
<td>N/A</td>
<td>N/A</td>
<td>0.14 (0.01) m</td>
<td>0.16 (0.02) m</td>
</tr>
</tbody>
</table>

Table 2. Mean (±SD) of the observed joint angles during the different testing sessions

<table>
<thead>
<tr>
<th>Variable</th>
<th>Session One Pre-Test</th>
<th>Session One Post-Test</th>
<th>Session Two Pre-Test</th>
<th>Session Two Post-Test</th>
</tr>
</thead>
<tbody>
<tr>
<td>Left Ankle Angle at Heel Strike</td>
<td>42.86 (5.15)°</td>
<td>43.86 (8.38)°</td>
<td>44.11 (7.23)°</td>
<td>40.79 (8.05)°</td>
</tr>
<tr>
<td>Left Knee Angle at Heel Strike</td>
<td>14.18 (8.21)°</td>
<td>13.17 (6.48)°</td>
<td>10.23 (10.31)°</td>
<td>9.62 (8.07)°</td>
</tr>
<tr>
<td>Left Hip Angle at Heel Strike</td>
<td>19.29 (5.64)°</td>
<td>20.11 (6.12)°</td>
<td>31.78 (9.90)°</td>
<td>33.18 (11.64)°</td>
</tr>
<tr>
<td>Left Ankle Angle at Toe Off</td>
<td>16.94 (4.27)°</td>
<td>16.79 (6.53)°</td>
<td>18.18 (10.68)°</td>
<td>28.48 (14.42)°</td>
</tr>
<tr>
<td>Left Knee Angle at Toe Off</td>
<td>5.31 (3.66)°</td>
<td>5.39 (2.42)°</td>
<td>18.18 (10.69)°</td>
<td>28.48 (14.41)°</td>
</tr>
<tr>
<td>Left Trunk Angle at Toe Off</td>
<td>5.31 (3.66)°</td>
<td>5.39 (2.43)°</td>
<td>12.20 (9.95)°</td>
<td>14.51 (10.26)°</td>
</tr>
<tr>
<td>Right Ankle Angle at Heel Strike</td>
<td>51.44 (12.17)°</td>
<td>50.50 (17.88)°</td>
<td>54.41 (14.12)°</td>
<td>46.16 (10.92)°</td>
</tr>
<tr>
<td>Right Knee Angle at Heel Strike</td>
<td>7.95 (3.69)°</td>
<td>11.09 (4.92)°</td>
<td>4.52 (3.80)°</td>
<td>11.03 (5.37)°</td>
</tr>
<tr>
<td>Right Hip Angle at Heel Strike</td>
<td>24.98 (10.90)°</td>
<td>24.49 (8.39)°</td>
<td>26.36 (9.67)°</td>
<td>18.76 (8.21)°</td>
</tr>
<tr>
<td>Right Ankle Angle at Toe Off</td>
<td>49.45 (19.69)°</td>
<td>47.94 (19.29)°</td>
<td>41.89 (15.92)°</td>
<td>38.35 (20.84)°</td>
</tr>
<tr>
<td>Right Knee Angle at Toe Off</td>
<td>28.26 (6.66)°</td>
<td>27.32 (7.49)°</td>
<td>17.58 (7.94)°</td>
<td>14.99 (8.13)°</td>
</tr>
<tr>
<td>Right Trunk Angle at Toe Off</td>
<td>4.95 (3.29)°</td>
<td>9.68 (9.27)°</td>
<td>3.21 (2.99)°</td>
<td>14.08 (12.14)°</td>
</tr>
</tbody>
</table>

There was a decrease in stride length for the 2010 riding program in post-riding measures as compared to the 2009 riding program pre and post-riding measures that would indicate a decrement in strength and stability as the condition progresses. One aspect of the normal progression of FA involves a reduction in the speed of walking (8). The indication is that maintenance of balance takes priority over locomotor propulsion. The current investigation did not measure the speed of walking; however the significant decrease in stride length is an indicator of balance and could indicate a progression of ataxia. Additionally, concurrent with slow walking speed is a small angular displacement of the hip, knee and ankle joints. The current study found no significant differences in joint angles from any of the pre- and post-riding measures. The lack of significant change in joint angle could indicate an interruption in the disease process.

DISCUSSION

Gait ataxia is usually the earliest symptom for individuals with FA (4). Muscle weakness of the hip
extensors and abductors are common and distal limb muscle weakness progresses with the advancement of FA. Gait disorders continue to progress until it is necessary for the individual to rely on a wheelchair for mobility, which occurs approximately 15 years after the onset of symptoms (7). Thus, slowing the progression of ataxia by maintaining proprioceptive awareness and muscle strength would indicate a delay in the progression of the symptoms of FA. Horseback riding has been suggested as an activity that stimulates proprioceptive awareness. Of particular interest in this case study was the observation of the rider’s posture during one of the more vigorous gaits of the horse. As the horse’s gait became more active, the rider’s posture dramatically improved.

When the rider's mount was required to trot, a two-beat gait involving diagonal pairs of legs, the riders sitting posture improved immediately without apparent effort. Riding a trot requires strong back and abdominal muscles, or postural muscles, because it jolts the rider due to an actual drop in the horses back when one diagonal pair of legs leave the ground before the other diagonal pair hit the ground. The sudden perturbation from the horses gait stimulated the riders postural righting reflexes, that is, the reflex activity used to maintain an upright posture. The stimulation of the righting reflexes caused an involuntary stressing of the postural muscles of the rider producing a training effect on the muscles. When the horse was at a walk, the rider had a very relaxed posture and appeared to be ready to slide out of the saddle at any given moment. Consequently, the same training effect would not be seen for the rider when the horse was at a walk rather than a trot.

More research is necessary to provide greater insight into the role physical activity and therapeutic horseback riding play on the management and locomotor skills of individuals with FA. Currently there is a lack of information to develop physical training and riding programs for individuals with FA, so recommendations are made using more common neuromuscular conditions. NARHA certified instructors generally follow the guidelines and recommendations for riders with MD when planning a program for a rider with FA. The rider with MD should not be over-stressed or fatigued because too much stress can hasten the degenerative process (5). The NARHA Standards and Accreditation Manual supports any attempt to encourage individuals with MD to remain active for as long as possible, however, indicates that fatigue is a precaution and could be a contraindication to participation in riding therapy (11). Therefore, NARHA recommends a shortened riding session for individuals with MD. Findings from the current study indicate that maintaining strength in the postural muscles by stimulating proprioceptive awareness through the righting reflexes help to delay the progression of ataxia. Currently, riders with degenerative muscle conditions would follow a riding program of 30 to 45 minutes once weekly. Implications from the current study indicate the recommendation for a therapeutic horseback riding program for an individual with FA should be conducted for at least 30 minute sessions, but increase the frequency to two or three times each week, and should include short intervals of trotting. Caution is advised, however, because this case study applies to one individual. More research is required to make generalized recommendations for physical training programs for individuals with FA.

Some of the limitations to the present study included measurement issues such as stride width. Stride width was only measured during session two, and therefore comparisons of stride width between the two riding sessions cannot be made. While Dartfish is a very useful motion analysis tool, it is limited in that it can only measure gait kinematics in two dimensions. A more sophisticated motion capture system would allow for the measurement of three dimensional gait kinematics, including motion in the transverse plane. In the present study, all data was collected at the horse park, so it was not feasible to set up a motion capture system that would require several cameras and optimal lighting. Future studies will examine the three dimensional gait kinematics as well as kinetics in a controlled laboratory setting. Finally, the participant had been involved with therapeutic horseback riding over the course of seven years, but only two years of data were collected for the current study because the program was ongoing upon investigator involvement. While the present study found encouraging results regarding the progression of FA in the participant, this may have been due to seven years of participation in the program and not just the two years that were examined.

In the present case study, the lack of significant changes in joint angles may indicate that therapeutic horseback riding helped prevent some of the decrements caused by FA. Future research studies should further examine if therapeutic horseback riding reduces or slows down the progression of ataxia among participants with FA, and how long these improvements in gait persevere. Additionally, more research needs to be done to evaluate the underlying reasons for the delay in ataxia for individuals with FA and if similar benefits may be obtained for individuals with other degenerative muscle conditions such as muscular dystrophy, multiple sclerosis, or amyotrophic lateral sclerosis.
REFERENCES


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