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Original Research

Three-Dimensional Kinematic Motion Analysis of Shivers in Horses: A Pilot Study

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ABSTRACT

Our aim was to assess three-dimensional kinematic motion analysis as an objective diagnostic tool for the characterization of the movement disorder of Shivers in horses. Kinematic parameters were measured in three horses with Shivers and were compared with a control group of four normal horses. Multiple parameter differences were found in the horses with Shivers at the walk, during backing, and when asked to pick up their hindlimbs. Most significant changes were a wider hindlimb stance of 0.39 ± 0.15 m and increased abduction angle of 48.7 ± 7.52° and hoof elevation (0.77 ± 0.08 m left and 0.94 ± 0.11 m right) when the horses were asked to pick up their hindlimbs. Control horses could back easily in a straight line and with symmetrical hoof separation and could maintain their center of weight when picking up their hindlimbs. In contrast, the horses with Shivers had difficulty backing straight, were slower, with a shorter stride and asymmetric hoof separation. They could not maintain their center of balance when picking up their hindlimbs. The findings of this pilot study advance the understanding of the movement disorder of Shivers and could be used as outcome measures to evaluate treatment modalities.

1. Introduction

Equine Shivers is a progressive movement disorder characterized by rapid hyperflexion and abduction of one or both hindlimbs with muscle trembling and elevation of the tail. These clinical signs are most seen when the horse is asked to pick up a hindlimb, when backed, or turned sharply and are exacerbated with transport, stress, and footing changes [1–4]. Once highly prevalent when horses were the mainstay of industrial work and transport, this condition diminished by the 1930's with the transition to automobiles and the mechanization of farming [5,6] but still remains as a significant disorder impacting the lives and performance careers of horses. In advanced stages, horses are severely affected, and despite the decades since it was first described, there is still no known treatment, with humane euthanasia as the only option for many horses. Research findings suggest that Shivers is caused by cerebellar Purkinje cell axonal degeneration, but there are many unanswered questions about its pathophysiology and need for an effective intervention [7].

Until recently, diagnosis of Shivers has been mainly subjective, based on presentation. Owners may be unaware that their horse has Shivers until the clinical signs are apparent. In some cases, it is difficult to diagnose whether a horse has Shivers or another movement disorder known as Stringhalt [3,4,8–11]. Stringhalt also causes an involuntary hyperflexion of one or both hindlimbs of horses, during backing and at the walk and trot. A kinematic study using semiquantitative analysis of the vertical displacement of the hindlimbs of affected horses in client videos identified differentiating features and subcategories of Shivers presentation [12]. Shivers, however, has not yet been evaluated with three-dimensional (3D) kinematic motion analysis. Three-dimensional

Animal welfare/ethical statement: This study was conducted solely by a private equine practice, DeClue Equine, LLC. All study horses were client-owned, with an established veterinarian-client-patient relationship with the primary investigator of the study (A.J.D.). The study was conducted following the guidelines outlined by the American Veterinary Medical Association's Principles of Veterinary Medical Ethics. Owners signed informed client consent and nondisclosure agreement forms before enrollment of their horse.

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kinematics has been a powerful modality to robustly characterize human and canine movement disorders [13–16]. It has been used to greatly advance the knowledge of equine locomotion of the distal forelimb and hindlimb joints of different gaits and breeds, over varying surfaces and alterations of hoof balance [17–23]. With the technology of 3D camera systems and noninvasive marker techniques, study of the biomechanics of the equine neck, back, and pelvis have also been explored, including the characterization of ataxia in horses [24–27]. We hypothesized that 3D kinematic assessment of horses with Shivers during backing and picking up their hindlimbs would identify additional key quantifiable parameters that would lead to a more complete understanding of this movement disorder.

The goal of this pilot study was to characterize Shivers in horses with 3D kinematic motion analysis. Fulfillment of this aim would identify differences between normal and Shivers-affected horses that would allow for reliable, objective parameters for accurate diagnosis and to evaluate the efficacy of treatment modalities for Shivers.

2. Materials and Methods

2.1. Horses

Seven horses were part of the pilot study (four normal healthy controls and three horses with Shivers). For inclusion into the study, animals were examined with a baseline assessment of their overall general health, including lameness and neurological examinations by the veterinary investigators of this study. Horses were evaluated for lameness at the walk and trot based on the American Association of Equine Practitioners lameness scale [28]. Horses were evaluated on a standard neurological examination for mentation, posture, cranial nerve deficits, cervicofacial reflex, cutaneous sensation, tail and anal tone, and evidence of gait abnormalities (ataxia, paresis, spasticity, and dysmetria) and proprioceptive deficits at the walk, during backing, circling, tail pull, and elevation of the head, and on different surfaces. The horses were graded for gait abnormalities with a modified Mayhew scale of 0 (normal) to 5 (recumbent) [29]. Before inclusion in the study, candidate horses with Shivers were evaluated by the study veterinarians based on the criteria for Shivers outlined in the recently published kinematic video study with evaluation of the horses walking forward and backward for at least 10 strides, turning sharply and manual lifting each limb. Shivers was defined by chronic difficulty backing characterized by either excessive hindlimb hyperflexion or excessive hindlimb rigidity or extension [12]. Horses with Shivers that had evidence of any underlying systemic illness, neurological deficits, and other gait abnormalities not primarily Shivers were not included in the study.

A detailed description of the study group horses is provided in a Supplemental Table (S2 Table. Study Horses). The control group consisted of 3 geldings and one mare between 9 and 14 years of age, with mean body weight of 511.4 ± 39.4 kg (ranging from 454.5 to 545.5 kg) and mean height of 158.8 ± 9.5 cm (ranging 145–165 cm). Horses were of varying disciplines (Hunter/Jumper, Dressage, Cutting) and breeds (Warmblood, Quarter Horse). All four horses were normal on lameness and neurological evaluation. The three horses with Shivers for the pilot study were all geldings between 4 and 14 years of age, with mean body weight of 450.8 ± 36.5 kg (ranging from 409.1 to 477.3 kg) and mean height of 147.6 ± 14.8 cm (ranging from 138.4 to 164.6 cm). Horses were of varying disciplines (Show, Western Pleasure) and breeds (American Saddlebred, Paint Horse, and Arabian) and had a history of bilateral Shivers >1 year. All 3 horses were not in work due to the severity of Shivers and their owners reported that the horses required sedation to be able to stand quietly for the farrier to trim their hindfeet.

2.2. Kinematic Analysis

Locomotion analysis was performed on the control horses and for the horses with Shivers at a private equine farm. Horses were acclimated initially with trial sessions to the digital video camera system and for optimization and validation of the study markers before collection of data for the kinematic study. Measurements were obtained with an infrared-based automated gait analysis system (OptiTrack, NaturalPoint, Inc, Corvallis, OR), operating at 240 Hz with 8 (Prime 13) cameras positioned for a measurement volume of 9.14 m × 12.2 m × 2.74 m (Fig. 1). Horses were groomed and then marked with 19 mm and 12.7 mm reflective spherical markers using double-sided carpet tape (Roberts, Boca Raton, FL) at 72 anatomical sites to define the lumbar, lumbosacral joint, pelvis, tail, stifle, tarsus, fetlock, and foot bilaterally (S1 Table) based on previously described equine marker locations and known anatomical landmarks [30–32]. All markers were applied to the horses by the same investigator to minimize the variability of marker placement that can occur with different human applicators [33]. Horses' tails were wrapped up in self-adhesive bandaging tape (3M VetRap, 3M, St. Paul, MN) to allow for marker visibility and to prevent markers from being inadvertently removed if a horse flicked its tail. Each session, photos of the horses were taken to record the duplicity of the marker placement, especially for the cluster markers (Fig. 2A). Measurements were taken initially with the horses standing square for 20 seconds for static calibration. Measurements were then taken with the horses walked for 10 strides, backed for 10 strides, and then with an alternating hindlimb leg lift sequence (minimum of three lifts per side). A minimum of three trials per gait were performed to ensure four useable strides (without the loss of markers). The same investigator handled the horses during the walking and backing sessions and when the horses were asked to pick up their hindlimbs (leg lift) again, to minimize variability of the horses' responses that could occur with different handlers. During each locomotive evaluation, the horses were also videoed simultaneously with 3 GoPro HERO5 Black (GoPro, Inc, San Mateo, CA) camera systems from the back and right and left sides of the measure volume.

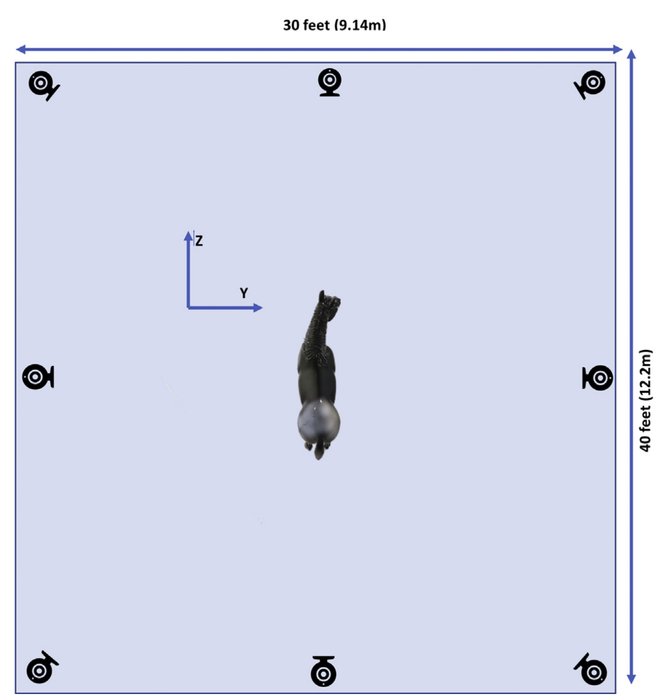
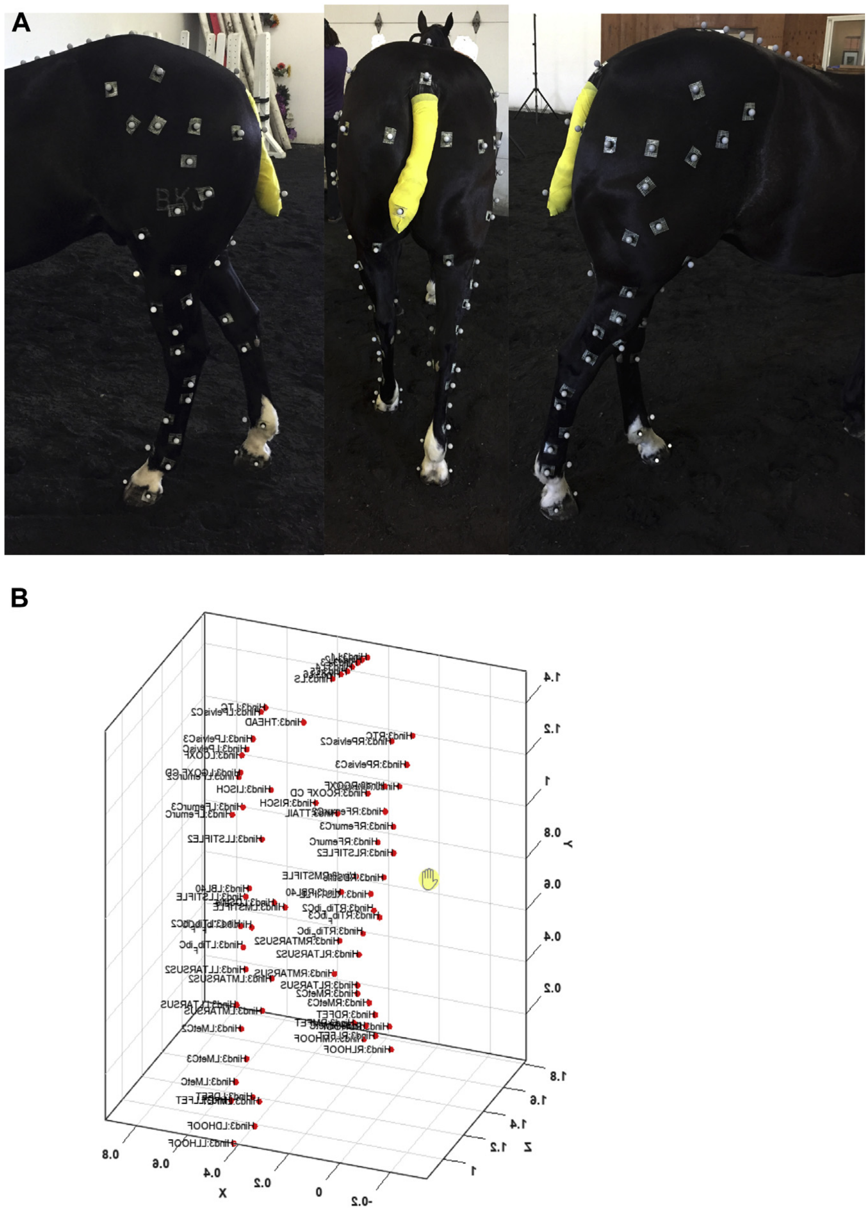


Fig. 1. Motion-capture video camera setup.



T. Secord

Fig. 2. Markers on horse and reconstruction of data coordinates for calculations. (A) Session photos of marker placements on study horse. (B) Reconstruction of raw marker data coordinates for biomechanical metrics with GNU Octave 4.4.1 software.

Reconstruction of the 3D position of each marker was based on a linear transformation algorithm (Motive: Tracker 1.2.2, NaturalPoint, Inc, Corvallis, OR). Raw coordinates were imported into the Visual 3Dv6 Professional (C-Motion, Research Biomechanics, Inc, Germantown, MD) analysis program for the evaluation of the coxofemoral, stifle, tarsus, and fetlock angles in flexion/extension, tail angle swing, and kinematic gait metrics (speed, step time, step length, stride length). The x-y-z Cardan rotational sequence was used for angular computations with the right-hand rule to determine polarity of angular variables. Kinematic data were filtered using a fourth-order Butterworth low-pass filter at a cutoff frequency of 6 Hz.

Additional biomechanical metrics to further characterize the horses with Shivers were performed with importation of the raw coordinates for analysis using a free software system GNU Octave 4.4.1 (www.octave.org) (Fig. 2B). Metrics evaluated included the abduction angle, centroidal motion, hoof elevation, hoof deviation (separation) from the midline, and pelvis angle. Each metric was computed directly using the spatial point data obtained with the video analysis system. The abduction angle was measured as the angle between a gravitationally aligned vertical reference line and the line formed by joining the centroid of the lumbar vertebral markers and the centroid of the right or left hoof markers. The overall centroid of the data points at each time step in the video was computed to provide an overall measure of gait smoothness (Fig. 3). Hoof elevation was measured as the gravitationally aligned distance between the right or left hoof centroid and the x-z plane (i.e., the ground plane established by the measurement system). Midline deviation of each hoof was measured as the normal distance in the x-z plane between the line of best fit to the lumbar points and the hoof centroid. Hoof separation was computed as the distance in the x-z plane between the hoof centroids (Fig. 4).

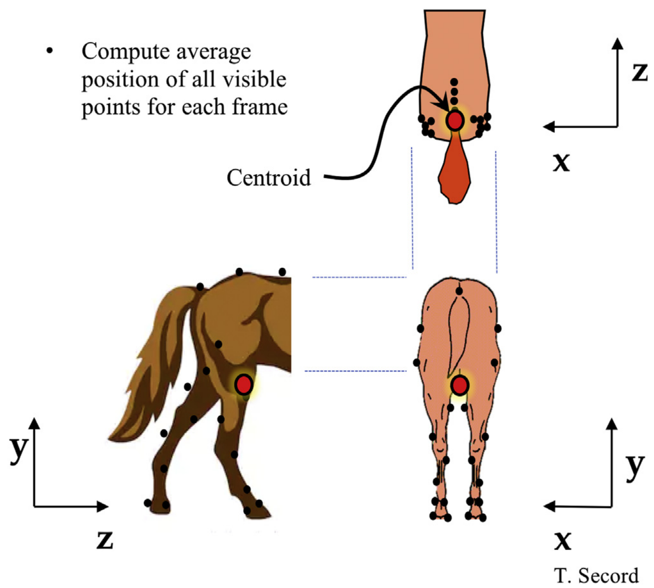


Fig. 3. Centroid position of hindlimb for backing calculations.

Finally, the pelvis angle was computed as the angle between the line formed by joining the centroids of the pelvis tracking points (COXF, COXF CD, and ISCH) and the ground plane (Fig. 5). Each metric was computed for all time frames in a video, although if insufficient data points were tracked in a given video frame, the metric was omitted at that particular video frame. Additional summary metrics that looked across the entire movement (e.g., maximum abduction angle) were computed without reference to null frames in the motion capture.

2.3. Statistical Analysis

Hindlimb joint angles, tail angle swing, kinematic gait metrics, maximum hindlimb abduction angle, centroidal motion (total distance, speed), maximum hoof elevation, maximum hoof deviation (separation) from the midline, and maximum pelvis angle at the walk and backing values were averaged across all recorded takes for each session for each horse. Mean values for horses with Shivers were compared against control horses with separate analysis of left and right hindlimb parameters. Statistical analysis for normality (Shapiro–Wilk) was performed for each mean data value, if normally distributed, a one-way repeated measures analysis of variance was performed with Bonferroni, and for non-normally distributed data, Brown-Forsythe method was performed. The level of significance was set at a P value of <.05. Statistical analysis was performed by using a computer spreadsheet statistical analysis package (SigmaPlot v.14; Systat Software, Inc, Point Richmond, CA).

2.4. Ethical Animal Research and Owner Informed Consent

This study was conducted solely by a private equine practice, DeClue Equine, LLC. All study horses were client-owned, with an established veterinarian-client-patient relationship with the primary investigator (A.J.D.) of the study. The study was conducted following the guidelines outlined by the American Veterinary Medical Association's Principles of Veterinary Medical Ethics. Owners signed informed client consent and nondisclosure agreement forms before enrollment of their horse.

3. Results

3.1. Standing

Spontaneous episodes of abduction and spasming of the hindlimbs for <5 seconds were observed in all 3 horses with Shivers. The horses had a significantly ($P < .05$) wider hindlimb stance compared with the control horses which stood with their tarsal joints and hind feet in line with the ilium (Table 1). One of the horses with Shivers stood with his hindlimbs out behind the vertical (“parked out”). In preparation for marker placement, all three horses were sensitive to being groomed, with the brushing provoking spontaneous Shivers.

3.2. Walking

The horses with Shivers were the least affected at the walk, but there were still noted differences which included a slower walking speed and mean stride length that was significantly ($P < .002$) shorter than the control horses (Table 1). Mild abduction and slight spasming of the hindlimbs were observed in the Shivers horses with in the initial forward movement of walking. The horses had higher and uneven hoof elevations, asymmetry of the left and right pelvis angles, and these were reflected in the hip, stifle, tarsal and fetlock joint angle kinematics (Tables 1 and 2). In contrast, the

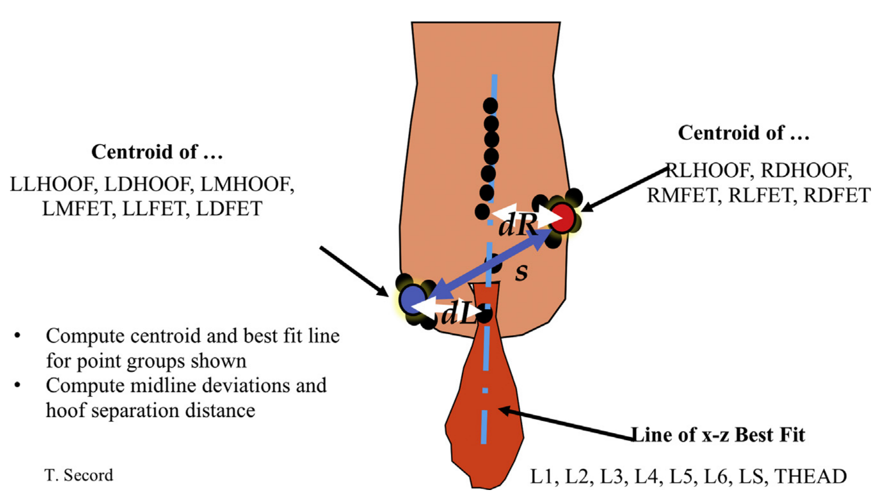


Fig. 4. Hoof separation and midline deviation calculation.

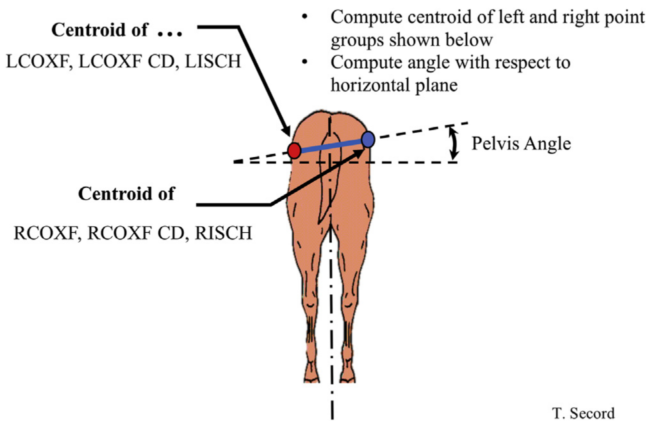


Fig. 5. Pelvis angle calculation.

control horses had more symmetrical step lengths, hoof elevations, and pelvis angles. The measured joint kinematic angles were comparable to previously reported values [23,30,34,35].

3.3. Backing

No studies characterizing the kinematics of horses when backed have been previously reported. The control horses had a backing speed of 0.62 ± 0.06 m/s, with a stride length of 1.14 ± 0.16 m and hoof elevation of 0.29 ± 0.03 m on the left and 0.25 ± 0.03 m on the right. Joint angle kinematics was comparable to walking values except for the hip joint flexion being narrower during backing (Table 3). The backing pelvis angles of the control horses were also symmetrical but at a slightly greater degree than during walking (Table 1). The control horses backed easily, without hesitation, and maintained a straight line of their lumbar midline with relatively symmetrical hoof separation (Fig. 6and S3 Video) and straight backing centroid (Fig. 7 and S4 Video).

The horses with Shivers were observed to be resistant to backing when asked and would brace their body and raise their head and neck up or down. The mean backing speed was slower in the Shivers horses and with a reduced stride length ($P < .05$) (Table 1). The hindlimb abduction and muscle spasming exhibited by these horses were reflected in the increased hoof elevations, asymmetric pelvis angles, and the corresponding increase in the hip, stifle, and tarsal joint flexion and fetlock joint extension (Tables 1 and 3). Unlike the control horses, the horses with Shivers were unable to back in a straight line. They would step laterally resulting in their lumbar midline traveling almost sideways rather than straight and with an asymmetric hoof separation from the midline (Fig. 6 and S5 Video). Their difficulties in backing were also apparent in the asymmetrical zigzag pattern of their backing centroid and increased distance traveled (Fig. 7 and S6 Video).

3.4. Leg Lift

The horses with Shivers were observed to have moderate to severe abduction and spasming of their hindlimbs <5 seconds and would shift their weight to the opposite limb or lose their balance (Fig. 8 and S7 Video) when asked to pick up their hindlimbs. The degree and quickness of the abduction was such that it was dangerous for the person asking the horses with Shivers, during the leg lift. In contrast, the control horses would pick up their hindlimbs without hesitation, easily, without moving away, and without shifting their weight (Fig. 8 and S8 Video). Specifically, the maximum abduction angle of the horses with Shivers was

Table 1
Kinematic differences.

	Control Horses	Shivers Horses
Standing		
Hindlimb Separation (m)	0.19 ± 0.09	0.39 ± 0.15*
Walking		
Walking speed (m/s)	1.21 ± 0.03	1.09 ± 0.30
Stride length (m)	1.72 ± 0.11	1.49 ± 0.15**
Step length (m)	0.84 ± 0.09(L)	0.78 ± 0.08(L)
	0.86 ± 0.06(R)	0.71 ± 0.11(R)**
Hoof elevation (m)	0.26 ± 0.04(L)	0.49 ± 0.35(L)
	0.26 ± 0.02(R)	0.42 ± 0.31(R)
Pelvis angle (°)	5.79 ± 3.15(L)	4.76 ± 4.43(L)
	6.76 ± 4.07(R)	12.9 ± 8.93(R)
Tail angle swing (°)	15.8 ± 16.2	35.1 ± 24.0
Backing		
Speed (m/s)	0.62 ± 0.06	0.51 ± 0.19
Stride length (m)	1.14 ± 0.16	0.90 ± 0.32*
Hoof elevation (m)	0.29 ± 0.03(L)	0.30 ± 0.06(L)
	0.25 ± 0.03(R)	0.66 ± 0.37(R)
Hoof separation (m)	0.44 ± 0.08(L)	0.39 ± 0.10(L)
	0.46 ± 0.06(R)	0.56 ± 0.19(R)
Pelvis angle (°)	8.83 ± 3.68(L)	7.13 ± 6.28(L)
	9.21 ± 2.32(R)	18.2 ± 12.9(R)
Tail angle swing (°)	7.63 ± 11.1	40.1 ± 41.3
Centroid distance (m)	17.9 ± 1.88	22.8 ± 7.54
Leg lift		
Abduction angle (°)	22.7 ± 3.53	48.7 ± 7.52*
Hoof elevation (m)	0.44 ± 0.08(L)	0.77 ± 0.08*(L)
	0.46 ± 0.08(R)	0.94 ± 0.11***(R)
Pelvis angle (°)	5.45 ± 2.84(L)	17.0 ± 10.0(L)
	5.75 ± 5.57(R)	27.0 ± 8.24(R)
Centroid distance (m)	13.5 ± 7.70	36.7 ± 23.5

Abbreviations: L, left hindlimb; R, right hindlimb.
Kinematic data: Mean ± SD.
*$P < .05$; **$P < .002$; ***$P = .003$.

significantly increased ($P < .05$) compared to the control horses (Table 1) with a significant difference ($P = .01$) in the abduction angle of the right hindlimb (mean = 37.4°) versus those in the control horses (mean = 31.1°). The maximum elevation of both the left and right hooves were also significantly increased ($P = .03$ and $P = .004$, respectively) in the Shivers horses compared to the normal control horses. The pelvis angles in the Shivers horses as they picked up their hindlimbs were not only asymmetrical but markedly increased from the control horses (Table 1) and reflected the greater degree of lateral shifting of balance from side to side as they picked up their hindlimbs (Fig. 8 and S7 Video).

4. Discussion

Three-dimensional (3D) kinematic motion analysis in this pilot study was successful in identifying additional multiple parameter differences in the horses with Shivers not previously described. The horses had a significantly wider hindlimb stance compared with the normal controls. The horses with Shivers had difficulty backing, characterized by a slower backing speed, shorter stride length, asymmetrical hoof elevation, and pelvis movement with a corresponding increased flexion of the hip, stifle, and tarsal joints and extension of the fetlock joints. When asked to pick up their hindlimbs, the horses with Shivers had significantly greater abduction and hoof elevation and asymmetry of pelvis movement.

Kinematic differences were also noted in the 3 horses with Shivers at the walk. These findings are contrary to the long-standing belief that Shivers does not affect the forward motion of horses as does Stringhalt. Recently, a previous kinematic study recognized a subcategory of more severely affected horses with Shivers that had increased vertical displacement and abduction of the hindlimbs on initiation of movement [12]. The differences in

Table 2
Walking joint kinematics.

			Control Horses			Shivers Horses		
			Mean ± SD	95% CI		Mean ± SD	95% CI	
Hip joint								
	Max flexion (°)	L	25.6 ± 3.65	23.0	28.2	35.7 ± 7.02	31.0	40.4
		R	28.0 ± 1.49	27.0	29.0	29.9 ± 6.18	26.0	33.8
	Max extension (°)	L	−2.46 ± 5.11	−6.12	1.20	5.97 ± 5.62	1.65	10.3
		R	−4.27 ± 7.34	−9.54	1.00	3.91 ± 7.80	−1.05	8.87
	Range of motion (°)	L	23.4 ± 12.2	19.9	26.9	31.0 ± 4.45	27.6	34.4
		R	30.4 ± 5.28	26.3	34.5	28.0 ± 6.07	23.7	32.4
Stifle joint								
	Max flexion (°)	L	69.0 ± 1.68	67.8	70.2	80.0 ± 10.8	72.7	87.3
		R	71.0 ± 5.83	66.8	75.2	77.0 ± 7.03	72.0	82.0
	Max extension (°)	L	37.6 ± 3.71	35.1	40.1	46.4 ± 8.07	40.2	52.6
		R	41.4 ± 8.89	34.6	48.2	39.7 ± 7.03	35.2	44.2
	Range of motion (°)	L	31.3 ± 4.57	28.0	34.6	34.5 ± 5.25	30.5	38.5
		R	28.1 ± 4.72	24.5	31.7	36.5 ± 5.57	32.5	40.5
Tarsal joint								
	Max flexion (°)	L	57.4 ± 4.31	54.3	60.5	77.4 ± 26.0	60.6	94.2
		R	55.5 ± 18.1	53.4	57.6	64.8 ± 10.6	57.7	71.9
	Max extension (°)	L	13.6 ± 5.14	10.2	17.1	22.1 ± 8.11	15.9	28.3
		R	13.1 ± 4.35	9.76	16.4	13.2 ± 6.68	8.95	17.5
	Range of motion (°)	L	43.5 ± 7.67	37.6	49.4	55.8 ± 21.2	39.5	72.1
		R	45.3 ± 4.22	41.9	48.7	53.3 ± 8.77	47.0	59.6
Fetlock joint								
	Max flexion (°)	L	33.7 ± 6.5	29.3	38.1	26.2 ± 4.12	23.0	29.4
		R	31.5 ± 4.33	28.4	34.6	32.5 ± 10.4	25.9	39.1
	Max extension (°)	L	−39.2 ± 9.13	−53.0	−25.4	−72.6 ± 30.0	−92.8	−52.4
		R	−48.4 ± 4.61	−51.49	−45.3	−51.6 ± 12.0	−65.6	−37.6
	Range of motion (°)	L	71.8 ± 10.8	63.5	80.1	98.1 ± 33.1	72.7	123.5
		R	78.7 ± 6.71	72.8	84.6	81.9 ± 20.0	67.6	96.2

Abbreviations: L, left hindlimb; R, right hindlimb.

walking speed, shorter stride length, asymmetry of the step length, and asymmetry in the pelvis angles of the horses with Shivers in this pilot study, however, were reflective of the overall measurements during the walk session and not only the initial steps. These were not clinically apparent to the human eye but were detectable with the more sensitive 3D digital camera system. The kinematic findings suggest that there may be additional changes not previously recognized in the forward gait of horses with Shivers.

Table 3
Backing joint kinematics.

			Control Horses			Shivers Horses		
			Mean ± SD	95% CI		Mean ± SD	95% CI	
Hip joint								
	Max flexion (°)	L	17.5 ± 4.47	15.0	19.97	26.3 ± 5.90	22.1	30.7
		R	20.8 ± 3.25	19.1	22.47	23.0 ± 7.40	17.7	28.3
	Max extension (°)	L	−2.23 ± 2.71	−3.58	−0.88	6.32 ± 3.85	3.87	8.77
		R	1.62 ± 4.04	−0.39	3.63	7.77 ± 6.32	3.75	11.8
	Range of motion (°)	L	19.6 ± 4.07	17.4	21.9	19.6 ± 5.19	15.7	23.5
		R	19.7 ± 4.99	17.1	22.3	14.6 ± 5.19	10.9	18.3
Stifle joint								
	Max Flexion (°)	L	82.0 ± 6.66	78.7	85.3	88.2 ± 7.99	83.1	93.3
		R	81.2 ± 4.66	79.0	83.5	101.9 ± 12.1	94.2	109.6
	Max extension (°)	L	36.3 ± 5.19	33.4	39.2	47.2 ± 8.80	40.9	53.5
		R	39.9 ± 4.05	37.8	42.0	39.5 ± 2.29	37.9	41.1
	Range of motion (°)	L	45.6 ± 5.52	42.5	48.7	42.1 ± 6.43	37.5	46.7
		R	40.9 ± 2.64	39.5	42.3	58.9 ± 8.60	52.8	65.1
Tarsal joint								
	Max flexion (°)	L	69.7 ± 23.9	56.5	82.9	80.2 ± 11.5	72.9	87.5
		R	66.3 ± 3.84	64.4	68.2	91.8 ± 17.8	80.5	103.1
	Max extension (°)	L	18.9 ± 5.35	15.9	21.9	29.2 ± 7.25	24.0	34.4
		R	21.4 ± 2.72	20.0	22.8	21.0 ± 1.76	19.7	22.3
	Range of motion (°)	L	47.6 ± 4.10	45.3	49.9	51.9 ± 7.13	46.8	57.0
		R	44.8 ± 4.19	42.7	47.0	65.7 ± 11.4	57.5	73.9
Fetlock joint								
	Max flexion (°)	L	39.6 ± 6.52	36.0	43.2	35.0 ± 2.83	32.7	37.0
		R	38.2 ± 6.54	34.8	41.6	31.1 ± 5.09	27.5	34.7
	Max extension (°)	L	−45.7 ± 11.2	−51.3	−40.1	−70.8 ± 17.1	−81.7	−59.9
		R	−52.0 ± 8.23	−56.1	−47.9	−71.4 ± 13.9	−80.2	−62.6
	Range of motion (°)	L	85.8 ± 12.1	81.5	90.1	106.3 ± 16.1	94.8	117.8
		R	89.8 ± 12.1	83.6	96.0	99.9 ± 13.1	90.6	109.3

Abbreviations: L, left hindlimb; R, right hindlimb.

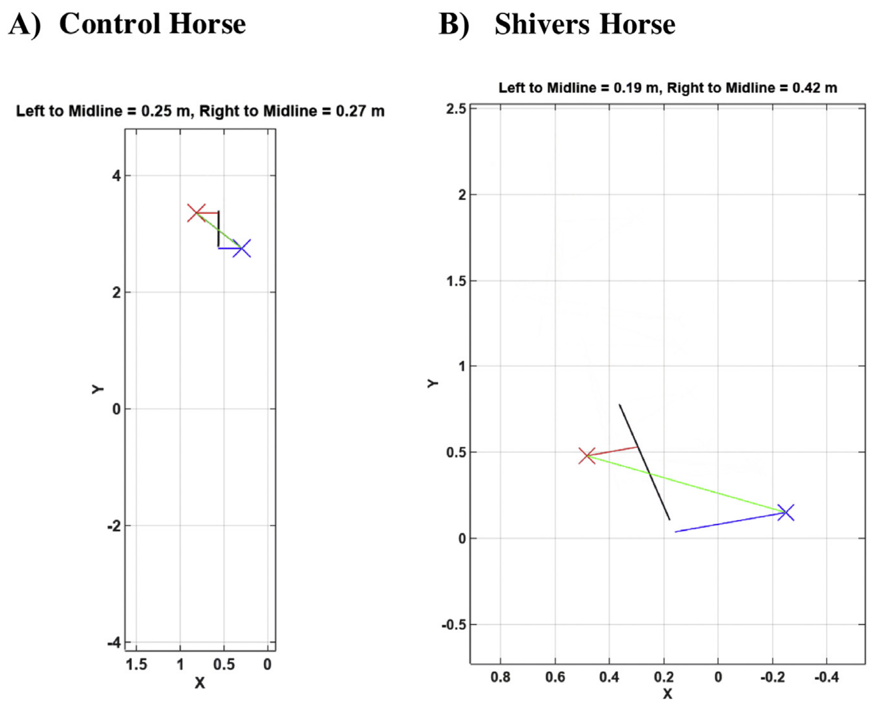


Fig. 6. Hoof separation backing. Still frame of animation video of the hoof separation of the horses when backed, viewed from above. Left hoof = red X; Left hoof distance from midline = red line; Right hoof = blue X; Right hoof distance from midline = blue line; lumbar midline = black line; green line = separation distance from left and right hindfeet. (A) Control horses backed in a straight line highlighted by the lumbar midline and maintaining an even separation of the right and left hindfeet. (B) The horses with Shivers had difficulty backing in a straight line and would step out more with one hindfoot as they backed, resulting in deviation of the lumbar midline. Full hoof separation video sequences of the control and Shivers horses can be viewed in the Supplemental Videos S3 and S5.

Evaluation of whether these differences persist at the trot would be important, as shivering horses are thought to have a normal trotting gait, unlike horses with Stringhalt [1,8,12]. Equally as illuminating would be a 3D kinematic study of Stringhalt in horses.

There were also observed, unquantified kinematic differences that were detected in the horses with Shivers beyond characterizing the hyperflexion and abduction of the hindlimbs. When asked to pick up their hindlimbs, the center of balance (visualized by the vertical midline) of the Shivers horses (S7 Video) was greatly deviated when compared with the control horses (S8 Video). Likewise, the backing videos of the control horses show clearly how their lumbar midline stays straight, between a more symmetrical hind hoof separation, compared to the Shivers horses (S3–S6 Videos). These differences combined with the increased and asymmetrical pelvis angles of the Shivers horses at the walk, during backing, and leg lift along with increased stance width, suggest that the lumbar and pelvis function are also affected. Differences in symmetry, shortening, and changes in speed, step, stride, and stance width, and changes in the center of gravity have been described in human patients with gait abnormalities due to back pain and pelvic dysfunction from instability of the core musculature, neurologic conditions, hip osteoarthritis, and tendinopathies [36–41]. As quadrupeds, horses are different from humans, however, further characterization of the kinematics of the pelvis and lumbar region of the horses with Shivers may provide insight into the underlying pathophysiology of this disorder.

One of the biggest challenges of the study was the application and maintenance of the markers on the horses. All three horses with Shivers in the study were highly sensitive to being groomed in preparation for the marker placements on the lumbar, pelvis, and hindlimbs. Two of the horses were extremely reactive to application of the markers on the lumbar region and on the lateral aspects of the gaskin and distal hindlimbs. Application and removal of the markers would provoke spontaneous Shivers episodes in the horses as they were being prepared, making it an adrenaline-fueled situation for the person handling the markers. Mapping of the cutaneous sensation responses of marker placements in the lumbar, pelvis, and hindlimbs of horses with Shivers in future studies could further illuminate potential etiologies for the hyperesthesia exhibited by these horses [42]. An additional observation noted in the horses with Shivers was the markedly increased degree of tail angle swing of the horses at the walk and during backing (Table 1) compared with the normal horses. Elevation of the tail has been described in horses with Shivers when backed but not at the walk [4,12]. Increased tail movement has been identified as a sign of musculoskeletal pain in horses in lameness evaluation [43]. Additional horses with Shivers will need to be evaluated to see if this increased tail movement is a consistent finding and could be used as a parameter to differentiate affected horses.

Three-dimensional kinematics has been a sensitive tool to characterize the muscle tremors and gait deficits associated with Parkinson's disease and the spasticity seen with cerebral palsy in humans, but the velocity and degree of movement of Shivers made acquisition of data for the complete 3D (frontal and transverse) joint kinematic analysis a challenge in the horses [13,14,44]. In addition, if a horse had an episode of Shivers during a video session, they would often slam down the foot hard enough that the markers would be shaken loose or fly off, resulting in having to start the video sequence over. Just turning a horse around in the camera setup for a sequence could exacerbate Shivers signs. Despite the difficulties that were faced, successful acquisition of complete and useable data sequences for the horses was achieved and we believe that 3D motion analysis is a viable and powerful modality to study Shivers in horses.

Data collection for the control horses was less challenging. The normal sensation responses and ease of movement made marker

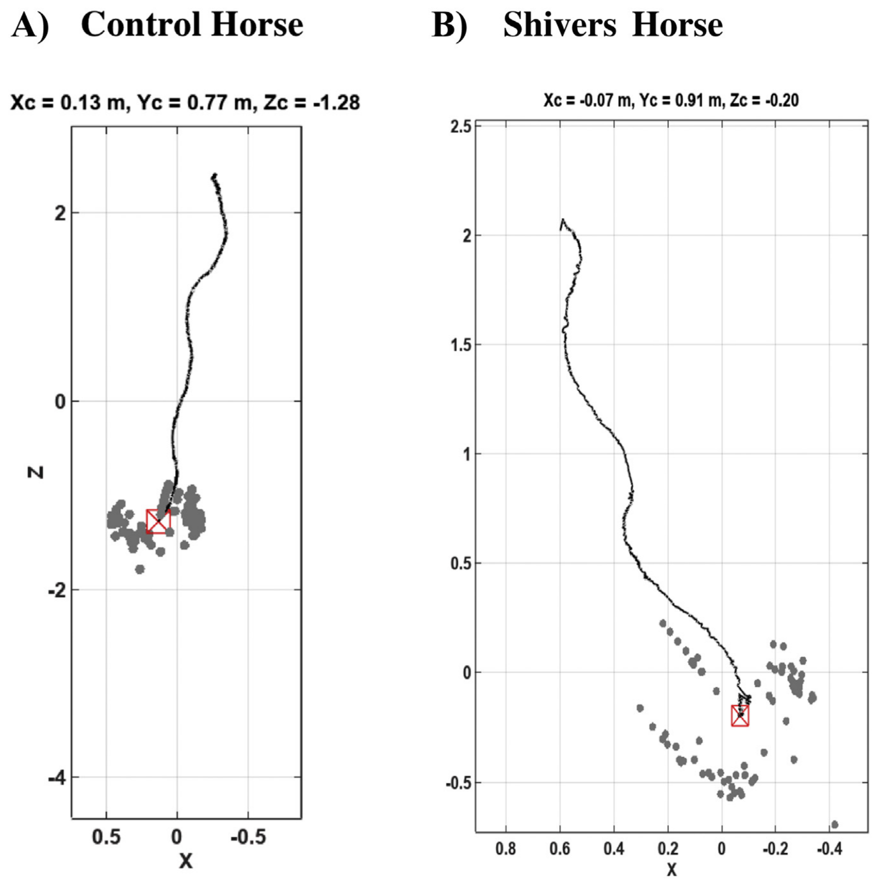


Fig. 7. Backing centroid. Still frame of animation video of the hind centroid position (red X box) of the horses when backed and viewed from above. (A) The tracking of the centroid position of the control horses remained relatively straight. (B) The horses with Shivers had a more zigzag tracking of the centroid deviating to the side to which the horse stepped more laterally. Full backing centroid video sequences of the control and Shivers horses can be viewed in the Supplemental Videos S4 and S6.

placement and maintenance in these horses relatively simple. The distal hindlimb joint kinematics of the tarsus and fetlock and metrics (speed, length, etc.) measured in the control horses were consistent with the results reported in other studies. It was noted, however, that in the control horses, although the range of motion measured for the hip and stifle joints at the walk were comparable with other reported studies [30,34,35], there were differences in the degree of flexion and extension for these joints. In a study by Back [34], a much greater degree of maximum flexion (93.7°) and extension of the hip (68.9°) were measured in horses walking at 1.6 m/second on a treadmill versus the values that were obtained for the controls (28.0° right hip flexion and −4.27° right hip extension) (Table 2). Likewise, Back reported a maximum stifle flexion angle of 46.1° and extension angle of 7.0° in his study, whereas in the control horses, a right stifle flexion angle of 71.0° and extension angle of 41.4° were measured. It is known that the high degree of skin displacement at the hip and stifle can have an impact in the variability of measurements in horses [34]. We also used different combinations than previously reported of markers to define the hip and stifle regions, to obtain centroids of the pelvis and hindlimbs for better characterization of Shivers. As an experimental consideration, further optimization of the marker sets for the pelvis and stifle used in future studies is indicated.

One major limitation of this study is the small number of animals, resulting in a potential lack of statistical power and in detecting differences. Another limitation is that exact size, breed, and age-matched controls were not used because of nature of this study being performed in private practice and as a pilot study. In a study comparing ponies to draft horses, size and body mass of horses were shown to correlate with an increase in the measured stride length of horses but a decrease in stride frequency [45]. Breed and confirmation-specific differences in gait characteristics has been described in Warmbloods, Andalusians, Tennessee Walking Horses, and Menorca horses [21,23,46,47]. Changes in fetlock joint angle were associated with increasing age in racing Thoroughbreds [48]. The availability at the time of recruitment of suitable Shivers cases and control client horses dictated the horses that were used in the pilot study, but an attempt was made to have a comparable study group (S2 Table. Study Horses). The horses used in the pilot study were all light breed horses ranging from 409.1 kg to 545.5 kg and height ranging from 138 cm to 165 cm. One of the horses with Shivers was 4 years old and had not started performing, but the remainder of the horses were middle-aged and had been or were performance horses. The 3 horses with Shivers were of different breeds (Paint, American Saddlebred, and Arabian) but were selected for their clinical severity of Shivers and were a true representation of the diversity of breeds/disciplines of horses with Shivers that the investigators (A.J.D. and K.K.S.) have seen in clinical practice. Nonetheless, in future clinical studies, matched controls might be more ideal to remove these as potentially confounding factors.

One final limitation was that the horses in this pilot study were more advanced cases of Shivers. It would be important to determine if the 3D kinematic changes detected are present in horses with milder presentations. For instance, the horses in this study did not exhibit the hyperextension of the front limbs seen in some

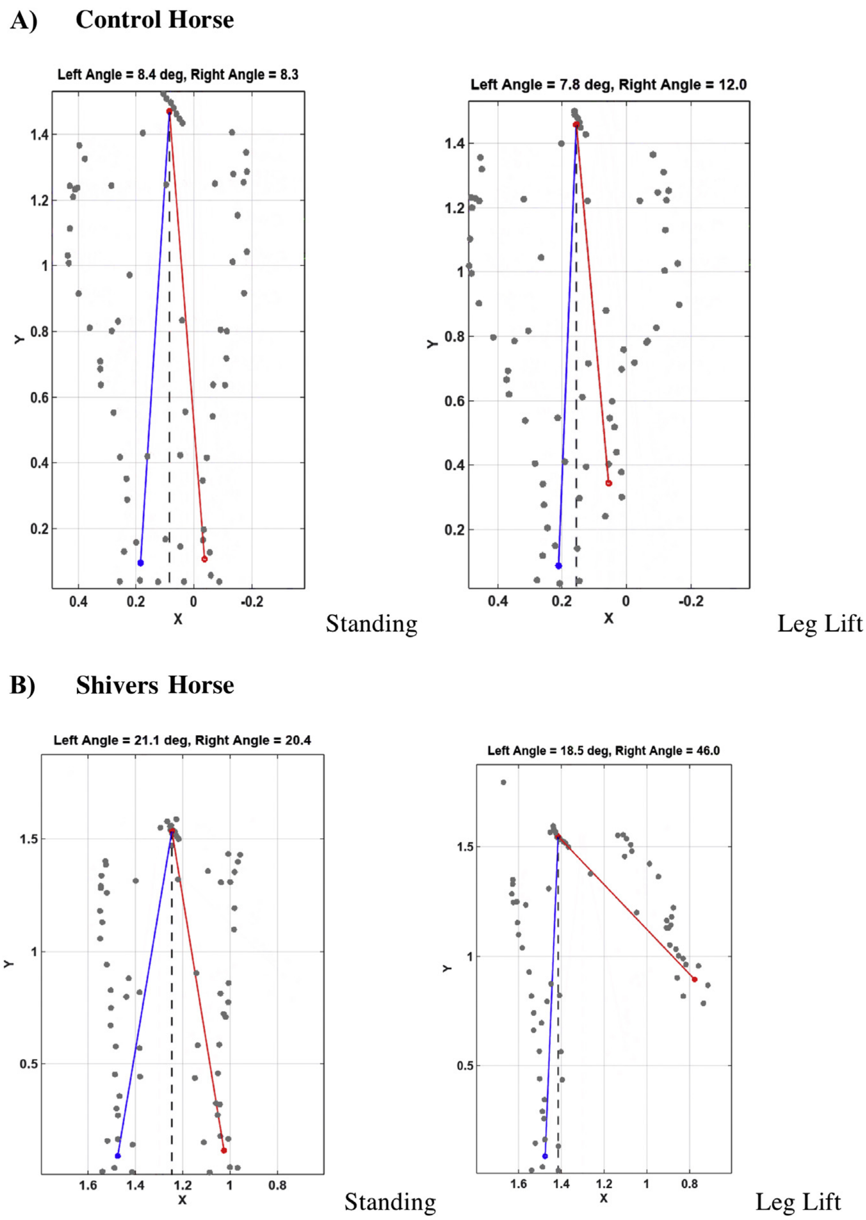


Fig. 8. Leg lift abduction angle. Still frame of animation video abduction angles of the left and right hindlimbs when horses were asked to pick up their hindlimbs. (A) The control horses would lift their hindlimbs maintaining their center of balance (vertical midline acting like a plumb line) with minimal angle of abduction during leg lift. (B) The horses with Shivers would either step to the side and/or deviation of the vertical midline to the opposite side with wide abduction angle of their hindlimb. Note the wide-based stance of the horses with Shivers versus the control horses. Full leg lift video sequences of the control and Shivers horses can be viewed in the Supplemental Videos S7 and S8.

horses with Shivers that were identified in a previous kinematic study. This subcategory of horses had vertical displacements of the hindlimbs that were not different from normal horses [12]. Further additional horses with varying degrees and different presentations of Shivers will need to be studied with 3D kinematics before a more complete characterization of this movement disorder in horses is achieved.

5. Conclusion

In conclusion, detectable differences in all three horses with Shivers in multiple parameters were measured with 3D kinematic motion analysis that were consistent with the clinical presentation of this movement disorder. The sensitivity of this modality allowed for detection of changes in the horses with Shivers at the walk and to characterize differences in the lumbar and pelvis function of these horses. Based on the preliminary results, we believe that this pilot study as an initial investigation demonstrates that 3D kinematic motion analysis is a sensitive and useful diagnostic tool that will advance the understanding of this movement disorder in horses. The study with the markers used and biomechanical analysis that was performed provides parameters to serve as objective outcome measures for future studies evaluating the response to treatment modalities.

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Authors' contributions: K.K.S. and A.J.D. designed the study. K.K.S., A.J.D., M.V., and S.K. were part of the data collection, handling of horses, and measurements. T.S. developed the additional algorithms, biomechanical analysis, and creation of animation videos with GNU Octave 4.4.1. K.K.S. and A.J.D. analyzed the data and K.K.S. performed the statistical analysis. All authors contributed to the writing of the article.

Supplementary Data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jevs.2019.03.006.

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