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Dynamic stability during level walking and obstacle crossing in persons with facioscapulohumeral muscular dystrophy

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ABSTRACT

Patients with FSHD suffer from progressive skeletal muscle weakness, which is associated with an elevated fall risk. To obtain insight into fall mechanisms in this patient group, we aimed to assess dynamic stability during level walking and obstacle crossing in patients at different disease stages. Ten patients with at least some lower extremity weakness were included, of whom six were classified as moderately affected and four as mildly affected. Ten healthy controls were also included. Level walking at comfortable speed was assessed, as well as crossing a 10 cm high wooden obstacle. We assessed forward and lateral dynamic stability, as well as spatiotemporal and kinematics variables. During level walking, the moderately affected group demonstrated a lower walking speed, which was accompanied by longer step times and smaller step lengths, yet dynamic stability was unaffected. When crossing the obstacle, however, the moderately affected patients demonstrated reduced forward stability margins during the trailing step, which was accompanied by an increased toe clearance and greater trunk and hip flexion. This suggests that during level walking, the patients effectively utilized compensatory strategies for maintaining dynamic stability, but that the moderately affected group lacked the capacity to fully compensate for the greater stability demands imposed by obstacle crossing, rendering them unable to maintain optimal stability levels. The present results highlight the difficulties that FSHD patients experience in performing this common activity of daily living and may help explain their propensity to fall in the forward direction.

1. Introduction

Facioscapulohumeral muscular dystrophy (FSHD) is a hereditary neuromuscular disorder leading to progressive muscle weakness. Skeletal muscle weakness leads to posture and balance impairments [1,2] and is related to a five times greater risk of recurrent falls [3]. Falls may lead to injuries but also to fear of falling, which causes a vicious circle of physical inactivity and secondary 'disuse' impacting muscle weakness, general health and quality of life [4].

Prevention of falls has been studied extensively in the elderly as well as in several groups of patients with neurological disorders (e.g. stroke, Parkinson's disease), but it has not yet been addressed in people with FSHD. The design of preventive intervention strategies may be informed by research on fall circumstances in people with FSHD. In this population, it was found that falls mainly occurred in the forward direction [3]. The predominance of forward falls seems counterintuitive, as in general the foot dorsiflexors and the abdominal muscles (that protect against backward falls) are affected at earlier disease stages and are also more severely affected than the calf and back muscles [5]. We previously demonstrated that, as a result of this muscle weakness, people with FSHD indeed have major difficulties sustaining balance perturbation in the backward direction as well. Hence, to better understand FSHD-specific fall mechanisms and for providing further directions for preventive strategies, insight is needed into the biomechanical challenges imposed on whole body dynamic stability during activities of daily living, such as walking and stepping over a doorstep.

Studies that applied quantitative, instrumented assessments of balance and gait in people with FSHD are yet scarce. A few studies on level walking have reported a decreased speed, step length and

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step frequency in persons with FSHD compared to healthy controls [6,7]. Furthermore, the ability of these patients to coordinate upper-body transversal plane movements during walking was found to become poorer with disease progression [8]. The defective upper-body control may compromise maintenance of dynamic stability. This interpretation is supported by a recent study on balance control following external perturbations induced by support-surface translations, in which we found that the degree of trunk muscle involvement associated strongly with postural instability in the sagittal plane [1]. It is yet unknown, however, whether people with FSHD indeed have poorer dynamic stability during self-initiated whole-body movements, such as walking, and in which stage of the disease these difficulties become manifest.

In the present study, we aimed to assess dynamic stability during walking in patients with various stages of FSHD. In addition, dynamic stability was further challenged by an obstacle that had to be crossed [9]. We hypothesized that compared to healthy controls, people with FSHD would demonstrate poorer dynamic stability during level walking. Defective dynamic stability was expected to become more apparent when crossing the obstacle and with more advanced stages of the disease. Furthermore, based on the previous report of frequent forward falls in FSHD, we expected greater impairments in the sagittal than in the frontal plane.

2. Methods

2.1. Participants

Ten patients with genetically confirmed FSHD were recruited via the rehabilitation and neurology departments of our university hospital. Patients with at least some weakness in pelvis or proximal legs were included (clinical severity scores (CSS) 3–4.5 as described by Ricci et al. [10]). They were categorized based on mild (CSS = 3) or moderate to severe pelvic and proximal leg muscle weakness (CSS $\geq$ 3.5) [6,7]. These two groups are further referred to as *mildly* and *moderately* affected. Specific exclusion criteria were the presence of other neurological diseases, severe cardio-pulmonary disease, contra-indications for MRI and pregnancy.

During an intake visit a physiatrist checked the inclusion and exclusion criteria and determined the CSS [10]. Upon inclusion, this visit was followed by an MRI scan at the same day. The methods and results of these MRIs have been reported elsewhere [11]. For the present study, we only used the average proportion of spared muscle tissue (PMT) to identify the most affected leg. Gait assessments were performed within 8 weeks after inclusion. A control group of similar age and gender distribution was also included. This study was approved by the local medical ethical committee. All subjects gave written informed consent.

2.2. Protocol

All subjects were asked to walk barefoot over a ten-meter walkway at their self-selected comfortable walking speed. Once subjects were accustomed to walking on the walkway, three trials of level walking were recorded. Subsequently, a wooden obstacle ($h \times l \times w$: 100 mm $\times$ 40 mm $\times$ 1000 mm) was placed in the middle of the walkway [12]. Subjects were instructed to step over the obstacle with both the left or right leg leading (three trials each in a random order). The starting position was at least 4 m in front of the obstacle to allow several steps to reach steady-state walking before the obstacle was encountered.

2.3. Data collection

Reflective markers were placed on the skin according to the PlugInGait full-body model (BodyBuilder, Vicon Motion Systems, Lake Forest, CA). Kinematic data were acquired using a six-camera Vicon motion analysis system (Vicon MX, Oxford Metrics, Oxford, UK) with a sample frequency of 100 Hz. Two steady-state gait cycles recorded from the middle portion of the walkway were used for analysis. Marker data was filtered (fourth-order Butterworth, 6 Hz) and processed with the Vicon Clinical Manager model (VCM) to calculate full body kinematics. Centre of mass (CoM) positions were calculated in the standard Vicon software as the weighted sum of the 12 body segments defined by the Plug-in-Gait model [13,14].

2.4. Data analysis

For both level walking and obstacle crossing, walking speed, step length, step width and step time were calculated from kinematic data. For obstacle trials, we calculated these variables for the pre-crossing step, the leading and the trailing step (Fig. 1). In addition, toe distance was calculated as the horizontal distance between the toe marker of the trailing leg (prior to crossing) and the obstacle. Heel distance was the horizontal distance between

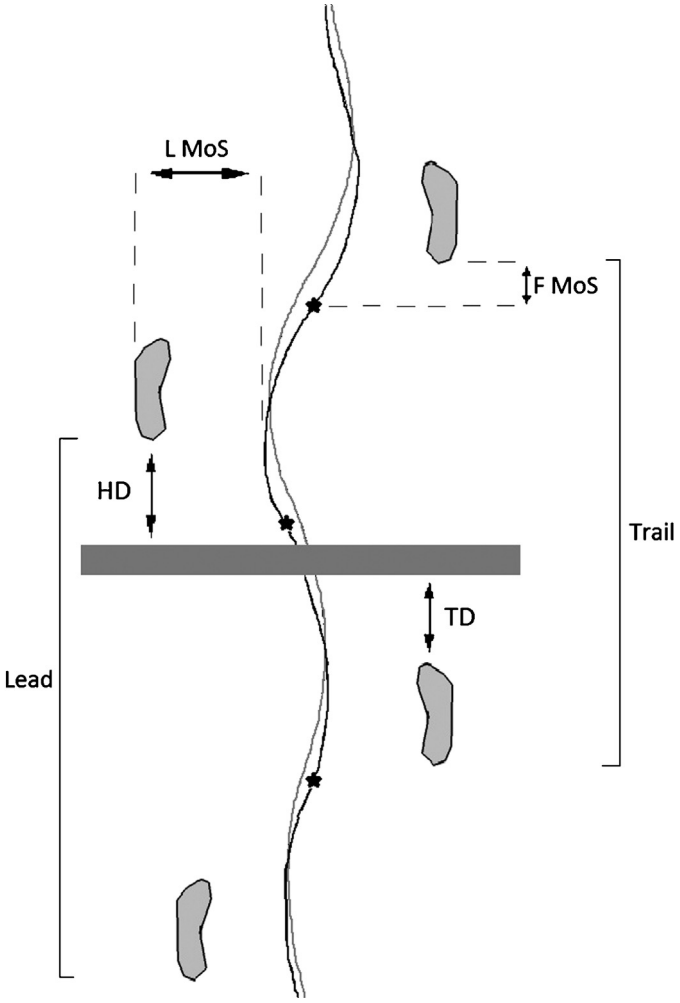

Fig. 1. Schematic overview of an obstructed walking trial displaying the leading step (Lead), and trailing step (Trail). The grey line represents the center of mass trajectory throughout the trial. The black line represents the extrapolated centre of mass (XCoM). Stars represent the moments of heel strike. Forward margin of stability (F MoS) is determined at heel strike. Lateral margin of stability (L MoS) reflects the smallest distance between XCoM and base of support throughout the stance phase. Toe distance (TD) is the distance between the toe marker and the obstacle. Heel distance (HD) is the distance between the heel marker and the obstacle, determined at foot contact of the leading limb.