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A systematic review of the gait characteristics associated with Cerebellar Ataxia

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ABSTRACT

Background: Cerebellar Ataxias are a group of gait disorders resulting from dysfunction of the cerebellum, commonly characterised by slowly progressing incoordination that manifests as problems with balance and walking leading to considerable disability. There is increasing acceptance of gait analysis techniques to quantify subtle gait characteristics that are unmeasurable by current clinical methods This systematic review aims to identify the gait characteristics able to differentiate between Cerebellar Ataxia and healthy controls.

Methods: Following systematic search and critical appraisal of the literature, gait data relating to preferred paced walking in Cerebellar Ataxia was extracted from 21 studies. A random-effect model meta-analysis was performed for 14 spatiotemporal parameters. Quality assessment was completed to detect risk of bias.

Results: There is strong evidence that compared with healthy controls, Cerebellar Ataxia patients walk with a reduced walking speed and cadence, reduced step length, stride length, and swing phase, increased walking base width, stride time, step time, stance phase and double limb support phase with increased variability of step length, stride length, and stride time.

Conclusion: The consensus description provided here, clarifies the gait pattern associated with ataxic gait disturbance in a large cohort of participants. High quality research and reporting is needed to explore specific genetic diagnoses and identify biomarkers for disease progression in order to develop well-evidenced clinical guidelines and interventions for Cerebellar Ataxia.

1. Introduction

Cerebellar Ataxias are a group of gait disorders resulting from dysfunction of the cerebellum and associated systems due to inherited and acquired causes. Cerebellar Ataxia (CA) is commonly characterised by slowly progressing incoordination which manifests as problems with balance and walking leading to considerable disability. Cerebellar Ataxias affect more than 10,000 adults in the UK [1], with variable age of onset and disease course.

Gait refers to the cyclic nature in which an individual walks, and is punctuated by consecutive heel strikes. An individual's body type, dictated by their sex, age and any natural physical asymmetries, affects their unique movement pattern [2]. Gait ataxia is clinically recognisable as a wide-based stance with truncal instability and irregular lurching steps, which can result in an increased risk of falls [3]. This can be accompanied or predominated by other symptoms depending on the ataxia subtype [4].

Presently, the principle methods of gait assessment in a clinical setting are through use of subjective rating scales such as the Scale for the Rating and Assessment of Ataxia (SARA) [5]. Although many of these are validated to detect progression of ataxia [6,7], there is evidence to suggest that clinical assessment scales might underestimate the severity of gait changes in CA [8].

Instrumented gait analysis techniques quantify subtle gait characteristics that would not be detected by clinical examination. There is increasing acceptance of the use of gait analysis methods such as 3D motion capture, pressure-sensitive walkways and inertial sensors for the assessment of neurological diseases that manifest with gait changes.

Improved classification of ataxic gait disturbance and definition of biomarkers for disease progression will enable quantification of the effect of novel and existing interventions to improve disease management in Cerebellar Ataxia while also clarifying the disease mechanisms in specific Cerebellar Ataxia subtypes [9].

Early studies using instrumented gait analysis in individuals with

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cerebellar syndromes described the spatiotemporal gait characteristics of Cerebellar Ataxia as: reduced cadence, step and length, gait velocity, and increased step and stride time and stance and swing phases [4,10]. However, other studies provide conflicting results and many report inconsistencies within cohorts. There are currently no guidelines to state the clinically relevant change in gait characteristics.

With technological advances making it quicker and easier to implement gait analysis, studies exploring neurological gait disorders are becoming more prevalent. It is now possible to seek a consensus description of the gait characteristics of Cerebellar Ataxia to explore the inconsistencies between published studies and to guide further research.

By evaluating and summarising the spatiotemporal gait characteristic measured using instrumented gait analysis techniques, this systematic review aims to answer the question: Which gait characteristics are able to differentiate between Cerebellar Ataxia and controls?

2. Methods

Available literature was systematically searched, following a predetermined protocol (PROSPERO 2016: CRD42016042149, Available from http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42016042149).

Using the PICOS framework [11] the research question was explored in order to guide design of search strategy and selection criteria. Of interest were studies where straight lined self-paced walking was measured in adults (age 18yrs or older) with Cerebellar Ataxia. Quantification of gait should involve instrumented techniques. Participants were not required to undergo any type of intervention as baseline gait characteristics were of most interest. Where healthy controls were recruited they should be matched for age and gender as a minimum. Studies of all designs were considered, except review articles, if published since 1996 and available in English.

2.1. Search strategy

The search strategy and selection criteria were developed in line with the review questions and agreed on by two researchers (AM, EB). Titles and abstracts of articles within a number of electronic databases (MEDLINE via OVID, psyc-INFO via OVID, PubMed, IEEE-xplore, Cochrane trials library, web of science core collections, and Scopus) were searched systematically implementing MESH search terms and key words where appropriate to combine three search phrases (walking terms (Walk* or gait or Locomotion); measurement terms (Measur* OR assess* OR evaluat* OR examin* OR analysis OR analy*e OR Biomechanic OR kinematic OR instrumented) and ataxia terms (Cerebellar Ataxia OR gait ataxia)) (Supplementary material 1). Searches were completed in July 2016; repeated in November 2016; and the output restricted to those published since 1996 until the search date. Reference lists from eligible articles as well as relevant reviews and systematic reviews were hand searched and studies identified subjected to the same selection criteria. This aimed to reduce any restrictions of the search strategy in uncovering unpublished and published evidence. Records identified were imported into EndNote (Clarivate Analytics); and processed to remove duplicate records and any older articles that remained.

2.2. Study screening process

Article screening was guided by an Inclusion/Exclusion criteria, predefined in line with the research question (Supplementary material 2). Titles and abstracts of articles identified by searches were subjected to the selection criteria by two researchers independently. References were divided between assessors in the interest of time, while 10% of articles were dual-screened to confirm appropriate decision-making and adherence to the selection criteria.

Those articles that satisfied the screening criteria moved on to full text appraisal. This was completed in parallel by assessors and final selections made through discussion. Where articles were suspected or confirmed to report results from identical or overlapping cohorts of patients the earliest or most relevant article was selected for inclusion.

2.3. Data extraction

Study information and gait parameters were extracted from the selected articles and, where necessary, authors contacted to request additional results. All available study information and reported gait characteristics was collated in Microsoft Excel.

Results were converted to common units of measurement, so that all spatial parameters were expressed in terms of metres (m) and temporal parameters expressed in terms of seconds (s). Speed was expressed as metres per second (m/s), cadence as number of steps in a minute (steps/ min) while phases of the gait cycle were expressed as a percentage of the total stride duration (%). Gait variability was reported as either Coefficient of Variation ((CV) defined as Standard Deviation (SD)/mean (%)) or combined Standard Deviation ((cSD) defined as the square root of the mean variance of the left and right steps (cm)) [12].

Where necessary, authors of selected articles were contacted to clarify study details and obtain unreported results. This included requesting mean average and standard deviation of cohort gait characteristics where median and interquartile range was reported and coefficient of variation where other variability measures (such as combined standard deviation) were reported. Articles where information was not made available for assessment following repeated requests were excluded from further analysis despite being potentially relevant studies. Where multiple subgroups were examined in a single study, data were combined to a single result following Cochrane Review guidance [11].

2.4. Data synthesis & meta-analysis

For cohort demographics, descriptive statistics (mean average, standard deviation (SD) and range) will be computed using IBM SPSS Statistics for Windows, version 23 (IBM Corp., Armonk, N.Y., USA)

Meta-analysis was completed in Rstudio (version 3.3.2) [13], using the “meta” package [14]. For parameters where results were available for more than 3 studies, the weighted mean difference (MD), 95% Confidence intervals (CI) and the standardised Z-score for overall effect were computed. Heterogeneity was tested using I^2 statistic, although a single group random effect model (REM) used throughout to give a conservative approach to meta-analysis.

Forest plots were generated to display the comparison of walking gait characteristics in Cerebellar Ataxia and healthy controls from preferred/comfortable self-paced walking.

Studies without control cohorts were included in the meta-analysis but not given any weighting in the calculation of the pooled estimate. To ensure the uniformity of data processing, gait parameters that had been standardised for individual biomechanical features (e.g. leg length or height), were excluded from meta-analyses. For gait variability, only coefficient of variation was reported commonly enough for results to be meta-analysed.

2.5. Quality assessment

Studies that were eligible for inclusion underwent quality assessment to detect risk of bias using an adaptation of the criteria described by Littell et al. [15] (Supplementary material 3). Researcher’s independent findings were compared, and ratings were agreed on through discussion.