**A survey of falls in people with dystonia.**

**Authors:** Melani J Boyce[1,2]*, Florence C.F. Chang[3], Neil Mahant[3], Victor S.C. Fung[3,4],

Lynley Bradnam[2,5]

**Affiliations:**

1 - Department of Physiotherapy, Westmead Hospital, Sydney, Australia

2 - Discipline of Physiotherapy, Graduate School of Health, University of Technology,

      Sydney, Australia

3 - Movement Disorders Unit, Department of Neurology, Westmead Hospital, Sydney,

      Australia

4 - Sydney Medical School, University of Sydney, Sydney, Australia

5 - Department of Exercise Science, Faculty of Science, University of Auckland, Auckland,

      New Zealand

**Corresponding Author:**

Melani Boyce
Department of Physiotherapy
Westmead Hospital,
Hawkesbury Rd,
Westmead, NSW, 2145
Australia
+61 2 8890 6500
Melani.boyce@health.nsw.gov.au

**Authorship:**

MB, LB – conception and design, acquisition and analysis of data, writing manuscript

FC, NM, VSCF – conception and design, writing manuscript

All authors have approved the final version of the manuscript

**Abstract**

*Objective*

Dystonia is a chronic and sometimes progressive neurological disorder causing abnormalities in movement and function. We conducted a preliminary survey to investigate whether people with dystonia experience falls and to identify contributing factors to falls in this population.

*Methods*

An online survey of people with dystonia was conducted in November 2015. Respondents were asked to complete demographic information, three questionnaires (the Falls Self-Efficacy Scale International [FES-I], the Activities-based Balance Confidence Scale [ABC] and the Functional Disability Questionnaire [FDQ]), and to report any falls sustained during the previous six months.

*Results*

Thirty nine percent of the 122 respondents reported falling in the previous 6 months and 65% of fallers were diagnosed with dystonia not affecting the lower limbs. Fallers reported lower falls self-efficacy and balance confidence with higher functional disability. Both falling

scales correlated with self-reported functional disability. Linear regression analysis for falls prediction revealed the variables FES-I and FDQ accounted for almost 30% of the falls in this dystonia population.

*Conclusion*

This survey indicates that fear of falling and balance confidence are impaired in people with dystonia, possibly impacting on function and falls. Further investigation into balance, function and falls in this population is required.

(196 words)

**Key words:** falls, physiotherapy, self-efficacy, survey, dystonia

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**Introduction**

Dystonia is a neurological disorder of motor control of uncertain aetiology affecting any or all the limbs, trunk or neck. It has been suggested that gait and balance are impaired in people with cervical dystonia (CD), as walking speed (Barr et al, 2017; Esposito et al, 2017), cadence, step length (Esposito et al, 2017), stepping reaction time and functional performance (assessed by the timed up and go [TUG]) are reduced in comparison to healthy controls (Barr et al, 2017; Hoffland et al, 2014). People with CD also experience reduced balance confidence and fear falling (Barr et al, 2017; Hoffland et al, 2014; Zetterberg et al, 2015), scoring significantly higher on the Falls self-Efficacy Scale International, (FES-I) than a healthy control group (Barr et al, 2017). However, it is unknown if walking impairments or fear of falling contribute to actual falls in people with cervical or other presentations of dystonia. The primary aim of this study was to establish whether people with dystonia have experienced falls and to identify contributing factors for falls in this population. Greater understanding of contributing factors to falls would lead to more effective physiotherapy interventions targeted at the physical and psychological impairments and functional limitations caused by dystonia.

**Methods**

A survey delivered by Qualtrics software was conducted over a 4-week period to internet support groups in Australia and New Zealand to explore the question as to whether people with dystonia have experienced falls and their potential contributing factors. The same data were used to validate the FES-I and the Activities Balance Confidence (ABC) scales in the dystonia population in an earlier publication (Boyce et al, 2017).

Following ethical approval (University of Technology Sydney Human Research Ethics Committee (UTS HREC ETH15-0096)) survey participants were asked to answer

demographic questions and to complete three surveys related to fear of falling and functional disability. The FES-I is a questionnaire designed to test a person's level of concern about falling during a range of common daily activities. The ABC is a questionnaire designed to test a person's confidence in their balance while performing common daily activities. The FES-I has excellent internal validity in the dystonia population (Cronbach's $\alpha = 0.97$) and a cut off value for predicting falls of 29.5 (Boyce et al, 2017). The ABC has high internal validity in the dystonia population (Cronbach's $\alpha = 0.97$) and a cut off value of 71.3 is predictive of a previous fall (Boyce et al, 2017). The Functional Disability Questionnaire (FDQ) determines the extent to which an individual's dystonia affects their engagement in daily tasks; a higher score indicates a higher level of functional disability. The FDQ has high internal consistency (Cronbach's $\alpha = 0.92$) (Jahanshahi and Marsden, 1990) and has strong concurrent validity with the fear of falling scales in the dystonia population (Boyce et al, 2017). Participants were asked if they had fallen over in the previous 6 months, and if so, were asked to record the number of falls and briefly describe circumstances surrounding them. For the purpose of this study, participants who reported one or more falls in the previous 6 months were considered to be "fallers".

Data were subjected to the Shapiro-Wilk test of normality, and then analysed descriptively to calculate means and standard deviations. Confidence intervals were calculated for the 3 questionnaires. Reported medication use was categorised into psychoactive or non-psychoactive by a qualified pharmacist. Participants were divided into a faller or non-faller group and each variable compared using independent student t-tests (2 tailed) for continuous data, and non-parametric Mann Whitney U comparisons for the questionnaires. Pearson's correlation coefficient was used to test for a relationship between the FES-I and ABC with the FDQ. Using falling as the dependent variable, a stepwise linear regression determined if any of the independent variables impacted on falling. These independent variables were:

FES-I, ABC, FDQ, age, gender, time since diagnosis, dystonia type, psychoactive

medications, botulinum toxin injections, and time since injections. Collinearity between

variables was assessed using the Variance Inflation Factor (VIF) during the linear regression

test. VIF values falling between 1-10 indicate little impact of multicollinearity. Statistical

analysis was performed using SPSS for Windows, Version 22 (SPSS, Inc., Chicago, IL,

USA). Statistical significance was set at $p < 0.05$.

**Results**

There were 122 responses to the survey. The characteristics of participants are available in a

previous publication by the authors (Boyce et al, 2017). The Shapiro-Wilk test indicated data

were normally distributed ($p > 0.05$). There were 48 (39%) self-reported fallers and 74 (61%)

non-fallers. Sixty-five percent of fallers were diagnosed with CD or dystonia of the face,

larynx or upper limb. The number of falls experienced by each type of dystonia is provided in

table 1. There was a difference between groups for the three questionnaires (all $p < 0. 018$),

with fallers reporting lower falls self-efficacy and balance confidence and higher functional

disability. The main reason for falling was loss of balance (such as a slip/trip or a fall) during

a functional activity. There was a difference between groups for dystonia type ($p = 0.033$),

with less CD in the faller compared to non-faller group. There was no difference between

groups for any other variables (all $p > 0.56$) (table 2). The Pearson's correlation coefficient

between the FES-I and FDQ was 0.77 and between the ABC and FDQ was 0.65 (both

p<0.001) indicating a moderately strong positive correlation between the two measures for

both falls questionnaires. Linear regression analysis revealed the variables FES-I and FDQ

together predicted falls (adjusted R-square =0.29, $p$ <0.0001). The ABC, age, sex, time since

diagnosis, botulinum toxin injections, time since injections or psychoactive medication did

not impact on falling (all $p > 0.17$). Collinearity VIP values indicated no multicollinearity,

although the moderate linear association between the FES-I and FDQ reported above indicates there may be collinearity between these two variables.

 [Insert table 1 here]

**Discussion**

The survey revealed that almost 40% of people with dystonia had experienced a fall, indicating that falling may be a significant issue for this neurological population. Falls self-efficacy and balance confidence were lower and self-reported functional disability were higher in the falls group. Low falls self-efficacy (FES-I) and higher disability levels (FDQ) were the only predictors of a fall from the variables collected. However, the FES-I and the FDQ were themselves moderately correlated, indicating only one scale may be required to predict falls in dystonia. The number of people with craniocervical dystonia who had experienced a fall was unexpected. Seventy-two percent of respondents experienced dystonia of the neck, face or upper limb and 65% of the fallers reported dystonia in these body areas. It is unclear why these people would fall, given their impairments are solely in the upper body or face, rather than segmental lower limb or generalised dystonia where the limb impairment itself could conceivably impair balance. The major reason for falling was loss of balance during functional activities or slipping/ tripping over external objects. It is possible that these falls may result directly from the impairments of dystonia (e.g. limb jerking and tremors); from the physical limitations of dystonia (e.g. restricted vision in blepharospasm), or other causes yet to be determined. Further study into the reasons why people with dystonia fall is required, particularly in light of their high fear of falling and low balance confidence. It is important that clinical trials investigating functional limitations and falling in dystonia include these questionnaires to compare psychological factors related to falling with the physical limitations imposed by the dystonia. Further studies should also examine whether

falls self-efficacy and balance confidence scales are predictive of impaired function measured by clinical functional scales (e.g. timed up and go, Berg Balance Scale) and laboratory testing of gait parameters and balance. Emerging evidence suggests an involvement of the cerebellum in the pathogenesis of dystonia (Jinnah and Hess, 2018; Prudente et al, 2014), which could more directly lead to falls given the cerebellum is integral to normal balance and lower limb coordination (Morton and Bastian, 2004). A possible dysfunction of the cerebellum leading to deficits in balance and falls remains to be explored in the dystonia population.

There are limitations to our survey. People who hold fall-related fears or had experienced a fall may have been more likely to complete the questionnaires and as the study was advertised online, people were indirectly excluded if they did not have access to the internet. Assessing falls retrospectively can be inaccurate in some populations and our results should be validated in a prospective falls study with a firm definition of what constitutes a fall (e.g. "an unexpected event in which the participant comes to rest on the ground, floor, or lower level", Lamb et al, 2005)). Finally, our regression analysis only included limited factors pertaining to falls risk, so the result should be interpreted conservatively until further research is undertaken on a wider range of dystonia and falls-related factors.

**Conclusion**

This study highlights that falls happen for people living with dystonia and that fear of falling and balance confidence are impaired in this population. People with dystonia who have fallen report a higher level of functional disability than those who have not fallen.  This raises important questions for future research investigating why and how people with dystonia fall and factors that may contribute to these falls. This understanding may lead to development and implementation of effective falls prevention interventions for people living with dystonia.

**Implications for Physiotherapy Practice**

- Thirty-nine percent of respondents reporting falling in the previous six months.

- Sixty-five percent of fallers had dystonia which did not affect their lower limbs.

- Fallers reported lower falls self-efficacy, less balance confidence and higher disability then those who did not fall.

- More research into balance and falling in people with dystonia is required so that physiotherapy interventions can be devised to address this problem.

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**Table 1. The survey population divided into Fallers and Non-Fallers**

| | Fallers N = 48 (39%) | | Non-Fallers N = 74 (61%) | | P value |
|---|---|---|---|---|---|
| Age (yr) | 51.2 ± 21.1 (range 21 – 74) | | 52.6 ± 11.6 (range 18 – 76) | | 0.56* |
| Gender | Male: 7 (15%) | | Male: 13 (17.6%) | | 0.67* |
| | Female: 41 (84%) | | Female: 61 (82.4%) | | |
| Time since diagnosis (yr) | 8 ± 7 (range 0.4 – 25) | | 9.8 ± 10.2 (range 0.5 – 53) | | 0.57* |
| Dystonia Type | CD: (45.8%) | 22 | CD: (61.4%) | 43 | 0.033*^ |
| | Segmental/focal UL: (6.3%) | 3 | Segmental/focal UL: (4.3%) | 3 | |
| | Segmental/focal LL: (1%) | 1 | Segmental/focal LL: (2.9%) | 2 | |
| | Facial/Blepharospasm: (4.2%) | 2 | Facial/Blepharospasm: (8.6%) | 6 | |
| | Generalized: (16.7%) | 8 | Generalized: (11.4%) | 8 | |
| | Multifocal: (18.8%) | 9 | Multi focal: (8.6%) | 6 | |
| | Spasmodic Dysphonia: (8.3%) | 4 | Spasmodic Dysphonia: (2.9%) | 2 | |
| Botulinum toxin injections | Yes: 27 (56.3%) | | Yes: 45 (60.8%) | | 0.62* |
| | No: 21 (43.7%) | | No: 29 (39.2%) | | |
| Psychoactive Medication | Yes: 28 (58.3%) | | Yes: 41 (55.4%) | | 0.89* |
| | No: 20 (41.7%) | | No: 33 (44.6%) | | |
| Number of falls (n) | Total falls: 155 | | Total falls: 0 | | |
| | 1 fall: 15 | | | | |
| | 2 falls: 17 | | | | |
| | 3 falls: 8 | | | | |
| | 4 or more: 8 | | | | |
| FDQ | 87.4 ± 19.9 (CI: 81.3- 93.5) | | 78.1 ± 15.1 (CI: 73.4- 82.8) | | 0.018#^ |
| FES-I | 39.5 ± 14.9 (CI: 34.9- 44.12) | | 26.4 ± 9.5 (CI: 23.4- 39.4) | | <0.0001#^ |
| ABC | 55.2 ± 27.6 (CI: 46.67- 63.29) | | 75.3 ± 22.1 (CI: 70.01- 80.51) | | <0.0001#^ |

FDQ – Functional Disability Questionnaire

FES-I – Falls Self-Efficacy Scale International

ABC – Activities specific Balance Confidence Scale

* Independent sample t-tests

# Mann Whitney U test

^ indicates significant result (p<0.05)