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Wheelchair Hockey Improves Quality of Life in People with Neuromuscular Disease

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Abstract

Background. Participation in sports is known to have positive effects on people's health and psycho-social well-being. Recently, physical activity implications for people with disabilities have been explored, showing promising results on quality of life and self-concept. However, few studies have specifically investigated the effects of participation in adaptive sports on neuromuscular patients' quality of life. **Objective.** To evaluate differences in psycho-social well-being between people affected by a neuromuscular disease who play wheelchair hockey and those who do not. Individuals playing an adaptive sport would report better quality of life, higher physical self-efficacy scores and more effective coping strategies, as assessed by self-reported measures. **Design.** Cross-sectional study. **Setting.** Data were collected during clinical follow-ups at the NEMO Clinical Center in Milan (Italy). **Participants.** A total of 25 patients affected by neuromuscular diseases, aged 18 to 40, participated in the study. **Main outcome measures.** The primary outcome was to compare quality of life between groups. Secondary outcomes were the comparisons of physical self-efficacy and coping strategies through self-reported measures. **Results.** Wheelchair hockey players scored significantly higher on the Quality of Life Index (specifically on the health/functioning and psychological/spiritual sub-scales) and reported better physical self-efficacy and perceived physical ability compared to the control group (i.e., patients who do not participate in any adaptive sport), controlling for age and pathology. On the contrary, no difference was found for coping strategies between the two groups. **Conclusions.** This study identified a significant association between participation in wheelchair hockey and improved physical and psychological well-being of people affected by neuromuscular diseases, compared to those who are not involved in adaptive sports.

Keywords:

Adaptive sports, adaptive athletes, Neuromuscular Diseases, Spinal Muscular Atrophy, Duchenne Muscular Dystrophy, quality of life, health status

1. Introduction

Neuromuscular diseases (NMDs) are chronic and progressive conditions comprising a heterogeneous number of pathological manifestations. Although they can vary in severity, these diseases cause great impairment in patients' mobility, muscle tone and strength, ultimately leading to a considerable loss of functional capacity and independence [1]. Standards of care have increased life expectancy even though an effective treatment reversing these conditions is yet to be found.

Thus, it is important to support and take care of patients' physical, psychological, and social well-being, according to a comprehensive definition of health. Based on World Health Organization (WHO) guidelines, the concept of Quality of Life (QoL) represents an important measure to assess how patients perceive themselves in relation to their position in life and in relation to their goals, expectations, standards, and concerns [2]. As such, QoL appears to be a subjective and dynamic aspect, which could be better understood through self-reported measures [3]. Studies on neuromuscular patients' quality of life show how social and psychological well-being are negatively affected by the progression of the disease [4, 5]. Currently the patients' quality of life is mainly improved through rehabilitation and physical therapy. However, these programs often lead to an excessive medicalization of patients, who are usually already over-exposed to hospital and care settings.

Recently, there has been interest in how adaptive sports could be complementary to rehabilitation, thanks to their therapeutic and recreational features [6-10]. Physical activities' benefits have been highlighted in both the general population [11] and among people affected by disabilities [12-15]. Physical activity can lower the risk of secondary health conditions (i.e. osteoporosis, heart disease,

diabetes and weight problems) [12], reduce psychological problems (i.e., depression and anxiety), improve well-being and self-perceptions (i.e., positive body image, quality of life, self-efficacy), and facilitate social contact and community reintegration by emphasizing the individuals' abilities rather than their limitations [12-16]. Nevertheless, few studies [9,11,17,18,28] have specifically investigated the effects of participation in adaptive sports on quality of life of patients affected by a neuromuscular disease.

Thus, the present study aims to bridge this gap by exploring the associations between participation in an adaptive sport—specifically wheelchair hockey—and physical and psychological well-being of people affected by NMDs. In wheelchair hockey, players with reduced muscle power play as a team using electric wheelchairs and a longer stick to control the hockey ball from a seated position. The current study focused on this adaptive sport because it is quite common in Italy and a vast group of patients referred to our clinical center is involved in this team sport. Specifically, in the Italian context, this sport started from an unofficial Wheelchair Hockey League set up by a group of athletes member of the Italian muscular dystrophy society (Unione Italiana Lotta alla Distrofia Muscolare, UILDM) and ended up in a national federation recognized by the Italian Paralympic Committee.

The main goal of the current investigation was to analyze quality of life differences between wheelchair hockey players and a cohort of individuals affected by NMDs who never played adaptive sports, considering the total score of the Quality of Life index as primary endpoint. It was hypothesized that individuals involved in wheelchair hockey would report a better quality of life. In addition, physical self-efficacy and coping strategies levels were assessed by means of self-reported measures in the two groups. Wheelchair hockey players were expected to display higher physical self-efficacy and more effective coping strategies compared to participants in the control group.

2. Material and methods

The current cross-sectional study involved participants affected by neuromuscular diseases—specifically Duchenne’s Muscular Dystrophy (DMD) and Spinal Muscular Atrophy (SMA). Although characterized by unique specificities, these are both chronic progressive disorders that cause proximal rather than distal weakness and may require, as a consequence, the use of a wheelchair [19,20].

The study protocol was submitted and approved by the local Ethical committee (reference number: 348-072019). Data collection was conducted between October and December 2019.

2.1 Participants

All eligible patients were consecutively enrolled during their clinical follow-up evaluations at the NEMO Clinical Center in Milan (Italy). Inclusion criteria were: clinically and genetically confirmed diagnosis of DMD or SMA; age between 18 and 40 years; wheelchair users; no cognitive impairment (equivalent score >1) as assessed using Raven’s Standard Progressive Matrices [21]. Exclusion criteria for recruitment were: tracheostomy; cardiac instability; pregnancy; participation in an adaptive sport other than wheelchair hockey. Clinicians assessed the physical criteria (i.e., diagnosis, age, cardiac and respiratory conditions) based on the clinical information available for each patient and approached the eligible subjects. All patients agreed to participate in the study and completed all the assessment measures.

The subjects were divided into two groups: Group 1 (sport group), involving patients playing wheelchair hockey (SG) and Group 2 (control group), involving patients not playing any adaptive sports (CG). Participants were enrolled in the study voluntarily and willingly, after signing an informed consent form.

2.2 Assessment tools

The following demographics and anthropometric measures were collected for each participant: age, gender, educational level, working status, weight, height, body mass index (BMI), pathological

condition, use of non-invasive ventilation (number of hours per day), drug treatment for emotional and/or sleeping problems, physiotherapy and/or rehabilitation program. The clinic's psychologist initially administered the Raven's Standard Progressive Matrices [21] to evaluate participants' cognitive conceptual and non-verbal functioning. Subjects with an equivalent score higher than 1 were included in the study and were asked by researchers (i.e., psychologists and physicians working at the Center) to fill out a paper-and-pencil questionnaire comprising several scales. If a participants' physical impairment prevented them from completing the questionnaire, a researcher did based on patient's verbal responses.

Quality of Life Index (QLI), by Ferrans and Powers [22] is an assessment tool used in several countries (including Italy) that applies to individuals with a wide range of disabilities. It consists of two parts, each comprising 33 items related to four domains: health and functioning, socioeconomic aspects, psychological/spiritual issues, and family context. The first part evaluates respondents' satisfaction for each item, while the second part assesses the importance attributed to each item. Both parts of the assessment tools are based on a 6-point Likert scale (from 1 "very dissatisfied/not important at all" to 6 "very satisfied/very important"). The total score and scores for each domain ranges from 0 to 30, with higher scores indicating a better quality of life. According to Ferrans and Powers' study [22,23], the mean score in the general population is 23 (S.D.=4.0), and a score less than or equal to 19 suggests a poor quality of life. A difference of 2 or 3 points is considered to be clinically significant. The QLI has shown good psychometric properties [23].

Physical Self-Efficacy (PSE) scale [24], Italian adaptation by Lupoli and De Lucia, is a 22-item scale that requires respondents to rate each domain on a 6-point Likert scale (from 1 "strongly agree" to 6 "strongly disagree"). Ten items account for the Perceived Physical Ability (PPA) sub-scale, while twelve items account for the Physical Self-Perception Confidence (PSPC) sub-scale. Up to date, there is no between patients' Minimal Clinically Important Difference (MCID) score for PSE scores.

Coping Orientation to the Problems Experienced – New Italian Version 25 (COPE-NVI-25), by Foà et al. [25], evaluates coping strategies adopted by patients to confront stressors and disrupting life events. Respondents are asked to rate how often they react in a certain way when confronting specific problems, using a 6-point Likert scale (from 1 “I do not usually do it” to 6 “I always do it”). There are 25 items, covering five coping strategies: social support, avoidance, positive attitude, problem-solving, self-transcendence. Up to date, there is no between patients’ MCID score for coping strategies’ levels.

2.3 Statistical analyses

Data were reported in text as median and interquartile range for continuous variables and number and percentage for categorical ones. All statistical analyses have been performed using SAS 9.3 (SAS Institute, Inc, Cary, NC) software. Shapiro-Wilk test and Levene test were used to test the normality of the distribution and the homogeneity of the variance, respectively. For baseline demographic and clinical features’ group comparison, the Fisher's exact test and the Mann–Whitney U test were used to determine statistical significance, as appropriate. Differences in quality of life were first assessed using Mann-Whitney U test. Then, the significant differences were also confirmed using a multivariable linear regression model adjusting by age and type of disease as covariates. Similarly, also physical self-efficacy and coping strategies between sport group and control group were first assessed using the Mann–Whitney U test, and the significant differences were then confirmed using a multivariable linear regression model adjusting by age and type of disease as covariates. All tests were two-tailed and a p-value < 0.05 was considered statistically significant.

3. Results

3.1 Sample description

For the present study, 33 participants were initially recruited (Figure 1 reports the recruitment flow-chart). Sport group did not include any female participants whereas the control group did. Since simple multivariable adjustment, we included only male subjects in both the sport and control group. Therefore, our final sample comprised 25 individuals. Table 1 summarizes descriptive analyses of the 25 enrolled patients.

The sport group (SG) comprised 15 participants (median age: 22.89 years [21.41 - 31.06]), while CG comprised 10 (median age: 24.73 years [22.65 – 30.27]). The two were well balanced regarding demographic and clinical characteristics. Overall, 11 participants were affected by DMD and 14 by SMA (11 type 2 and 3 type 3 SMA).

3.2 Comparative univariate analysis

Univariate comparisons are reported in Table 2.

Participants who practice wheelchair hockey reported an overall significantly higher QLI score compared to the control group (21.70 [18.77 – 24.26] vs 16.68 [15.50 – 18.55], $p=0.0071$; Table 2). Additionally, scores on the health/functioning and the psychological/spiritual sub-scales significantly differed between groups: participants in the study group scored significantly higher than those in the control group (21.71 [17.23 – 24.73] vs 15.57 [14.73 – 16.08], $p=0.0051$ for health and functioning sub-scale; and 23.71 [18.92 – 26.07] vs 16.40 [15.86 – 18.00], $p=0.0030$ for psychological/spiritual sub-scale; Table 2). No significant difference among groups was found on the socioeconomic and family sub-scales of QLI.

Participants in the SG showed a higher total Physical Self-Efficacy (PSE) score and reported higher scores on the Perceived Physical Ability (PPA) sub-scale compared to the CG (3.70 [3.29 – 4.04] vs 3.12 [2.83 – 3.35], $p=0.0051$ for PSA; and 3.20 [2.80 – 3.70] vs 2.00 [1.60 – 2.80], $p=0.0097$ for PPA; Table 2), while no difference was found in the Physical Self-Perception Confidence (PSPC) sub-scale.

As for the Coping Orientation to the Problems Experienced scale, wheelchair hockey players and the control group did not significantly differ on any of the five coping strategies assessed by the questionnaire COPE-NVI-25.

3.3 Comparative multivariate analysis

When accounting for age and pathological condition, differences between study and control groups on quality of life and physical self-efficacy scales hold significance, as shown in Table 2. In detail, when considering the transition from the CG to the SG, an increase equal to 5.79 points (2.03 – 9.54) was estimated for the health and functioning sub-scale ($p=0.0042$), an increase equal to 5.74 points (1.72 – 9.77) was estimated for the psychological/spiritual sub-scale ($p=0.0074$), and an increase equal to 4.44 points (1.28 – 7.60) was estimated for the QLI total score ($p=0.0081$). Moreover, regarding the physical self-efficacy scale, the transition from the CG to the SG was associated to an estimated increase of 0.89 points (0.23 – 1.55) in the PPA sub-scale ($p=0.0104$), and to an estimated increase of 0.68 points (0.26 – 1.10) in the PSE total score ($p=0.0030$).

4. Discussion

The present research explored associations between adaptive sports participation and several measures of physical and socio-psychological well-being, with the ultimate goals of bridging the literature gap concerning athletes affected by NMDs and shedding light on possible positive effects of adaptive sports. These findings showed that people affected by NMDs playing wheelchair hockey have a significantly better quality of life than those who do not participate in adaptive sports, consistent with our hypothesis. Interestingly, the difference between the two groups in terms of quality of life index total score, which was the primary outcome measure, reached both statistical and clinical significance. Additionally, the study group reported a significantly higher physical self-efficacy score in comparison to the control group. These results are in line with previous findings

[14] and highlight an association between participation in sports and improvements in psychological problems (i.e., anxiety and depression), body image, self-efficacy, self-competence, and quality of life [18]. However contrary to previous investigations, the current study explored a more homogeneous population of neuromuscular patients (all affected by DMD or SMA) and analyzed the impact of wheelchair hockey participation on well-being.

In a study by Yazicioglu et al. [6], people with disability involved in an adaptive sport reported higher quality of life levels in physical, psychological, and social domains. The present findings were in line with results regarding the physical (the QLI's "health/functioning" domain) and psychological (the QLI's "psychological/spiritual" domain) areas. Several other studies involving people with different disabilities and chronic conditions showed that those involved in adaptive sports or following a specific exercise program reported better quality of life [28,29] and higher satisfaction with their life [30]. Côte-Leclerc et al. [26] also found that people with a disability playing an adaptive sport had quality of life scores comparable to those of the overall normative population.

In the current study, sport participants did not significantly differ from controls on the family sub-scale of Quality of Life. This result is consistent with Côte-Leclerc et al. [26] and Groff et al. [27] studies, which showed that participants with physical limitations playing adaptive sports scored significantly lower on the family QLI's sub-scale and on the family life and family participation domains respectively, compared to participants from the general population. Highly involved professional adaptive athletes reported difficulties in balancing sport and family time [28], which might explain current and previous findings. On the other hand, these results could simply highlight the fact that physical activity has a positive impact on the individual aspects (e.g., increased self-esteem, improved general health and well-being perception) rather than on relational ones. Further studies should explore the possible reasons that might explain the lack of satisfaction in family life among people who perform adaptive sports.

This study showed that wheelchair hockey players report a better perception of their physical ability compared to equally impaired patients who are not involved in any kind of adaptive sport. High scores on the Perceived Physical Ability (PPA) sub-scale subsequently drove higher physical self-efficacy overall scores. In line with Bandura's cognitive theory [32] and with Gill's findings [34] in the general population, perceived self-efficacy could foster the decision to initiate a specific sport activity, but could also help maintain the activity over time. As a key element to support individuals' health, the understanding of the associations between physical self-efficacy and self-concept of people with disabilities should be a primary topic for future investigations in this area. On the other hand, no correlations were found between sport activity and the Physical Self-Perception Confidence (PSPC) sub-scale. To our knowledge, previous research suggested that sport activity has a positive impact on self-concept and exercise self-efficacy in children and adolescents with disabilities [11,31] and in the general population [32]. In this case, the lack of correlations between sport activity and the Physical Self-Perception Confidence (PSPC) sub-scale appeared to be consistent with the characteristics of the population involved in the study: neuromuscular diseases are chronic and progressive and affect the patients' physical appearance in a specific and unfortunately irreversible way, that could reduce physical aspect confidence.

Regarding coping strategies, previous studies have explored the relationship between coping strategies adopted to confront challenging sport-related situations and effects and involvement in sport activity, both in the general population [35,36] and with adaptive athletes [37,38]. Originally it was hypothesized that participants involved in an adaptive sport could face unique challenges and booster their cognitive appraisal and behavioral reactions, gaining as a result the acquisition of effective coping strategies (i.e., problem-solving, positive attitude, social support). Nevertheless, results showed no significant difference between the two groups.

Despite the novelty of the present investigation, this study has some limitations. First, sample size was small, and might have hindered the statistical power. Moreover, only male individuals were

included in both the study and control groups to avoid confounding effects related to differences in gender distribution among the two groups. Prevalence of male subjects in the sport group might reflect the gender disparity in sport's contexts, an issue that has also been raised in the healthy population [39]. This, however, might have impaired generalizability of the current results. Further, the present research adopted a cross-sectional design, which did not allow causal inferences. Indeed, it was not possible to understand whether better quality of life and higher self-efficacy were determined by participation in adaptive sports, or rather participants with higher levels of quality of life and self-efficacy were more likely to participate in such activities. Experimental and longitudinal studies could be more effective in detecting this causal relation and should thus be pursued for further understanding the topic.

Additionally, the focus on wheelchair hockey players inevitably narrowed results generalizability. Being a team sport activity, wheelchair hockey might have uniquely contributed to improving self-perception of psychological well-being and emotional adjustment because it allows participants to consolidate friendships and positive relationships with fellow teammates. Thus, future research could benefit from including a wider range of both individual and team activities to shed light on how the type of adaptive sport might shape participants' self-perceptions. Moreover, length of involvement in the adaptive sport activity and reasons for playing wheelchair hockey were not assessed in the current study. However, this information could provide a more comprehensive understanding on the relations between sport participation and quality of life of people with disability.

5. Conclusions

The present study highlights that participants playing wheelchair hockey report significantly better quality of life and higher physical self-efficacy scores compared to those who do not play adaptive sports. These findings suggest the importance of sport activities especially adaptive ones (e.g.,

wheelchair hockey) as part of the patients' care intervention. While the current examination highlights adaptive sports' benefits on quality of life, self-perceptions, and psychological well-being, future studies should additionally explore advantages of these activities on patients' physical features (i.e., fatigue, fitness, mobility) for a more comprehensive understanding of adaptive sports' impact on individuals with neuromuscular disorders.

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Table 1. Demographics and clinical characteristics of our sample				
	Overall	SG	CG	p-value
Sample size	25	15	10	.
Age	24.55 [21.58 – 30.27]	22.89 [21.41 31.06]	24.73 [22.65 – 30.27]	0.8461
Disease, n (%)				0.4132
DMD	11 (44.00)	6 (40.00)	5 (50.00)	
SMA Type 2	11 (44.00)	8 (53.33)	3 (30.00)	
SMA Type 3	3 (12.00)	1 (6.67)	2 (20.00)	
NIV, n (%)				0.6265
Yes	21 (84.00)	12 (80.00)	9 (90.00)	
No	4 (16.00)	3 (20.00)	1 (10.00)	
Educational level (yrs), n (%)				0.8414
8	1 (4.00)	1 (6.67)	0 (0.00)	
13	20 (80.00)	12 (80.00)	8 (80.00)	
16	2 (8.00)	1 (6.67)	1 (10.00)	
18	2 (8.00)	1 (6.67)	1 (10.00)	
PT, n (%)				0.4028
Yes	15 (62.50)	10 (71.43)	5 (50.00)	
No	9 (37.50)	4 (28.57)	5 (50.00)	
Missing	1 (.%)	1 (.%)	0 (.%)	
Drug use, n (%)				0.2268
Yes	1 (4.17)	0 (0.00)	1 (10.00)	
No	23 (95.83)	14 (100.00)	9 (90.00)	
Missing	1 (.%)	1 (.%)	0 (.%)	

Abbreviations: SG, Sport Group; CG, Control Group; DMD, Duchenne Muscular Dystrophy; SMA, Spinal Muscular Atrophy; NIV, Non-Invasive Ventilation; PT, Physiotherapy.

Data are expressed as median [interquartile range], except where otherwise indicated

Bold values denote statistical significance at the $p < 0.05$ level.

Table 2. Univariate and multivariable (*) comparisons between the two analyzed groups.

	SG (n=15)	CG (n=10)	<i>p-value</i>	<i>B (95% ci)*</i>	<i>p-value*</i>
Quality of Life Index					
Health and functioning	21.71 [17.23 – 24.73]	15.57 [14.73 – 16.08]	0.0051	5.79 (2.03 – 9.54)	0.0042
Socioeconomic	21.21 [18.29 – 23.75]	16.69 [15.57 – 21.00]	0.1830	.	.
Psychological/spiritual	23.71 [18.92 – 26.07]	16.40 [15.86 – 18.00]	0.0030	5.74 (1.72 – 9.77)	0.0074
Family	24.00 [20.00 – 30.00]	21.25 [16.80 – 24.00]	0.1254	.	.
QLI Total score	21.70 [18.77 – 24.26]	16.68 [15.50 – 18.55]	0.0071	4.44 (1.28 – 7.60)	0.0081
Physical Self-Efficacy					
PPA	3.20 [2.80 – 3.70]	2.00 [1.60 – 2.80]	0.0097	0.89 (0.23 – 1.55)	0.0104
PSPC	4.17 [3.83 – 4.67]	3.88 [3.08 – 4.50]	0.1737	.	.
PSE Total score	3.70 [3.29 – 4.04]	3.12 [2.83 – 3.35]	0.0051	0.68 (0.26 – 1.10)	0.0030
COPE-NVI-25					
Social support	21.00 [15.00 – 23.00]	17.50 [15.00 – 21.00]	0.2007	.	.
Avoidance	10.00 [8.00 – 12.00]	10.00 [8.00 – 12.00]	0.5192	.	.
Positive attitude	32.00 [31.00 – 36.00]	32.00 [31.00 – 36.00]	0.9777	.	.
Problem-solving	23.00 [19.00 – 25.00]	23.00 [19.00 – 25.00]	0.6348	.	.
Self-transcendence	6.00 [5.00 – 14.00]	6.00 [5.00 – 14.00]	0.6504	.	.
COPE-NVI-25 Total score	92.00 [90.00 – 98.00]	92.00 [90.00 – 98.00]	0.4696	.	.

Abbreviations: SG, Sport Group; CG, Control Group; ci, confidence interval; QLI, Quality of life index; PPA, Perceived Physical Ability; PSPC, Physical Self-Perception Confidence; PSE, Physical Self-Efficacy; COPE-NVI-25, Coping Orientation to the Problems Experienced – New Italian Version 25.

All data are expressed as median [interquartile range].

Bold values denote statistical significance at the $p < 0.05$ level.

*Multivariable analyses adjusted for the effect of age and type of disease.

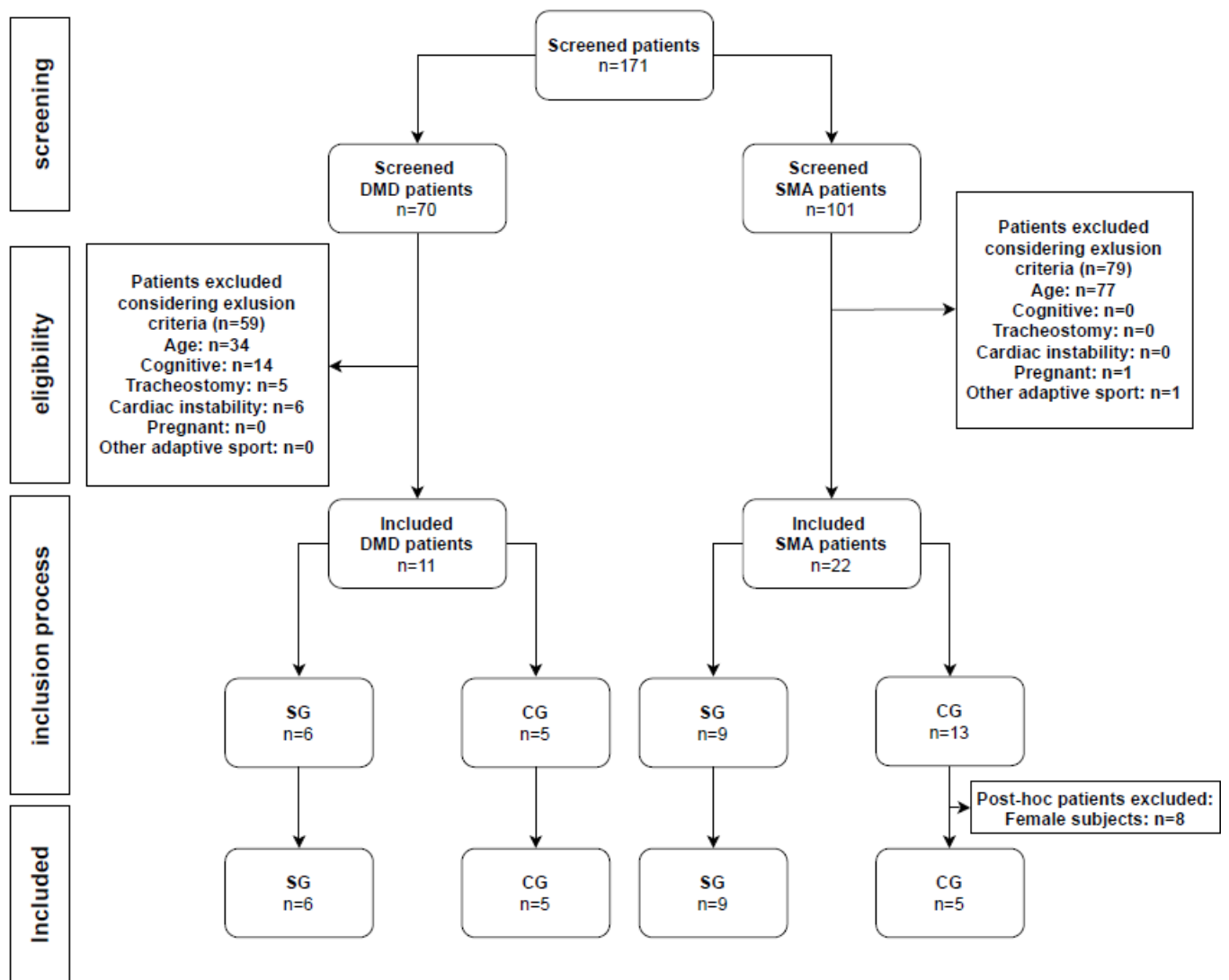


Figure Legend

Figure 1. Flow-chart diagram reporting the process of patients' enrollment in the study, based on inclusion and exclusion criteria.