

# Effectiveness of Teleassistance at Improving Quality of Life in People with Neuromuscular Diseases

Oscar Martínez<sup>1</sup>, Amaia Jometón<sup>1</sup>, Manuel Pérez<sup>1</sup>, Esther Lázaro<sup>1</sup>, Imanol Amayra<sup>1</sup>,  
Juan Francisco López-Paz<sup>1</sup>, Mireia Oliva<sup>1</sup>, Pamela Parada<sup>1</sup>, Sarah Berrocoso<sup>1</sup>, Andrea Iglesias<sup>1</sup>,  
Patricia Caballero<sup>1</sup>, Luis De Nicolás y Martínez<sup>1</sup> and José Eulalio Bárcena<sup>2</sup>

<sup>1</sup> Universidad de Deusto (Spain)

<sup>2</sup> Hospital de Cruces (Spain)

**Abstract.** Rare neuromuscular diseases (NDs) are a group of inherited or acquired neurological pathologies affecting the muscles and the nervous system. Their low prevalence and high geographical dispersion can cause isolation and difficulties in social interaction between affected equals. New technologies, such as videoconferencing, offer a complementary option for improving the health of this population. The purpose of this study was to assess the effectiveness of a teleassistance program at improving health-related quality of life (HRQoL) through social interaction in adults with NDs. The sample consisted of 45 participants affected by rare NDs. Twenty-four participants were assigned to the experimental group (EG), which participated in the videoconferencing sessions, and 21 to the control group. Three questionnaires were administered: WHO-DAS II, Sickness Impact Profile, and SF-36 Health Survey. Effectiveness was assessed by a pre-post design. An online psychosocial program was applied over three-month period. Data revealed an improvement of the EG in psychosocial variables, e.g. "Getting along with people" ( $z = -2.289$ ,  $r = -.47$ ,  $p \leq .05$ ) or "Psychosocial Domain" ( $z = -2.404$ ,  $r = -.49$ ,  $p \leq .05$ ), and in physical variables, e.g. "Life activities" ( $z = -2.844$ ,  $r = -.58$ ,  $p \leq .05$ ). Social interaction appeared as a relevant factor at improving HRQoL levels. High levels of satisfaction about the teleassistance program were reported.

Received 16 May 2013; Revised 27 February 2014; Accepted 13 May 2014

**Keywords:** teleassistance, videoconferencing, neuromuscular disease, quality of life, social interaction.

Rare Neuromuscular diseases (NDs) are different genetic illnesses with a chronic course of disease progression, low prevalence and high geographical dispersion. The main symptoms of these disorders are progressive loss of muscular strength, atrophy or hypertrophy, fatigue, muscular weakness, and spasticity or rigidity. Muscles or nerves are affected, and other organs and tissues, such as the heart or the central nervous system, may also be involved (ASEM, 2008).

Illness-related symptoms can cause high levels of disability in people with rare NDs and can worsen health-related quality of life (HRQoL). It is important to address the difficulties these populations experience in their daily routines to improve their general health status and their quality of life. Several studies have examined HRQoL in people with rare NDs. Tworck, Wiesmeth, Klewer, Pöhlau, and Kugler (2010) indicated lower levels of HRQoL in a sample of 1,518 patients affected with Myasthenia Gravis (MG) compared to those in a healthy group. A negative correlation between illness stability and mental health was

also found. Otherwise, Leonardi et al. (2009) pointed out that HRQoL of the MG patients studied in their study was inversely proportional to the illness gravity. Similar decrease in HRQoL was found in other studies with other type of disorders: amyotrophic lateral sclerosis, facioscapulohumeral muscular dystrophy (FSHD), and other dystrophies (Kierkegaard, Harms-Ringdahl, Holmqvist, & Tollbäck, 2011; Winter et al., 2010). Previous studies have also reported that psychosocial dimensions, such as social support and self-efficacy, are relevant in the improvement of HRQoL (Fioravanti, Casale, Mantegazza, Leonardi, & Raggi, 2010). HRQoL and psychosocial variables can be improved using new technologies.

The concept of telecare or teleassistance, meaning the use of new technologies to provide health care services, includes a wide range of modalities such as videoconferencing, e-mail, online chat sessions, forums, telephone calls, and mobile phone messages. These tools could be considered an alternative, innovative way to improve the HRQoL of people with disabilities (Ferrer-Roca, Garcia-Nogales, & Peláez, 2010; Lázaro, Amayra, López, De la Cruz, & Alday, 2009; Polisen et al., 2010).

Teleassistance has been used in the setting of multiple diseases, including cancer (Kroenke et al., 2010),

---

Correspondence concerning this article should be addressed to Oscar Martínez, Faculty of Psychology and Education, Universidad de Deusto, Avenida de las Universidades, 24. 48007. Bilbao (Spain).  
E-mail: oscar.martinez@deusto.es

diabetes (Polisena et al., 2009), post-traumatic stress disorder (Sloan, Gallagher, Feinstein, Lee, & Pruneau, 2011), obesity (Watson, Bickmore, Cange, Kulshreshtha, & Kvedar, 2012), chronic respiratory failure (Vitacca et al., 2009), cystic fibrosis (Hubbard, Broome, & Antia, 2005), brain injury (Rotondi, Sinkule, & Spring, 2005), chronic pain (Macea, Gajos, Daglia Calil, & Fregni, 2010), and stroke (Chumbler et al., 2010). The majority of these studies consider teleassistance a beneficial and useful resource.

Few studies have examined the application of new technologies to improve health in people with rare NDs (Amayra, Lázaro, López, & De la Cruz, 2008; Lázaro et al., 2009; López, Amayra, Lázaro, De la Cruz, & Alday, 2009; Soutter et al., 2004). These studies conclude that teleassistance is an important and useful tool at improving quality of life of children and adolescents with Duchenne muscular dystrophy, along with their relatives. Furthermore, Vitacca et al. (2010) conducted a study on telecare in patients with amyotrophic lateral sclerosis (ALS) and their caregivers and concluded that the applied program could be useful for following up with ALS patients. Likewise, the study of Boeschoten et al. (2012) found that online applied problem solving therapy is feasible and reduces depressive symptoms in patients with multiple sclerosis.

Health-related quality of life can be affected by a lack of social networks or by daily problems coping with a disease. Mobility limitations or the challenges this population may face in locating other rare ND patients nearby can be palliated by videoconferencing. This tool is an available way of keeping in contact with people with rare NDs who live in rural areas and may not have easy access to psychosocial care services. Rural living and the elevated rate of geographical dispersion of this population can complicate the creation of social networks among equals. Studies about this topic conclude that telemedicine or teleassistance are suitable methods for assisting people with rare NDs in rural environments (Ferrer-Roca et al., 2010; Rabinowitz et al., 2010) or geographically isolated populations (Backhaus et al., 2012). Furthermore, these authors highlight that videoconferencing psychotherapy “*is a viable alternative to in person therapy*”.

Satisfaction with teleassistance has been assessed in some studies as a measure of the acceptance of this new tool of intervention. Mair and Whitten (2000), for example, conducted a review of 32 studies on videoconferencing use and concluded that teleconsultation is an acceptable method of connection in a variety of circumstances. Doorenbos et al. (2010) found that online support groups are a valuable method of connecting people who have similar experiences. Another study, conducted by Gustke, Balch, West, and Rogers (2000), showed that participants’ satisfaction

with 495 interactive teleconsultations in real time was high (98.3%). Finally, Vitacca et al. (2010) found that 79% of patients and caregivers were highly satisfied with the telemedicine program they used.

The purpose of the present study responds to the lack of actual researches about teleassistance and HRQoL in people with rare NDs. The aim is to assess the effectiveness of an online psychosocial support program in this population to improve HRQoL levels through social interaction with affected equals.

## Methods

### Participants

Forty-five people (23 males and 22 females) with MG, FSHD, Becker muscular dystrophy (BMD), limb-girdle muscular dystrophy (LGMD), and other related diseases constituted the sample. The patients were recruited from the Hospital of Cruces and the Hospital of Basurto (Biscay), as well as from national associations of people affected by NDs. The participants’ ages ranged from 19 to 79 (mean = 50.56, *SD* = 14.07) years. A convenience sampling was conducted, assigning participants to one of two groups: the Experimental Group (EG) (*n* = 24), which received the online intervention, or the Control Group (CG) (*n* = 21). Bearing in mind the low prevalence of the population under study and the difficulty in obtaining the sample, the availability of a computer and a webcam was considered a main variable when assigning a person to the experimental group. Eligibility criteria included: confirmed ND diagnosis by a neurologist (ICD-10), age greater than 18 years, and agreement to participate in the study as demonstrated by signing an informed consent form. Participants in the EG were also expected to have a computer, a web camera, and access to the Internet at their homes. Exclusion criteria included severe psychopathology (e.g., psychosis) and/or sensorial difficulties that affect the management of the teleassistance program.

### Instruments

A structured interview was administered at the beginning of the study to assess participants’ sociodemographic information. HRQoL was measured using the World Health Organization Disability Assessment Schedule II (WHO-DAS II) (World Health Organization, 2001), the SF-36 Health Survey questionnaire (Ware & Sherbourne, 1992), and the Sickness Impact Profile (SIP) (Bergner, Bobbitt, Pollard, Martin, & Gilson, 1976). Previous studies have investigated the suitability of these instruments for assessing quality of life levels, and satisfactory results have been obtained (Leonardi et al., 2010; Winter et al., 2010).

The WHO-DAS II measures disability. The 36-item version was administered in this study. Participants were asked to indicate on a 5-point scale (from 1 "no difficulty" to 5 "complete difficulty/cannot do") if they have had any difficulty in the last 30 days due to health status in performing a variety of tasks related to 6 different domains: 1) Understanding and communicating, related to concentration, memory, problem solving, learning new tasks, and communication; 2) Getting around, referred to the difficulty in standing for periods of 30 minutes, standing up from sitting down, moving inside home or getting out of one's home, and walking long distances; 3) Self-care, which includes activities such as getting dressed, washing the whole body, eating, and staying by oneself for a few days; 4) Getting along with people, which includes aspects related to maintaining a friendship, interpersonal skills with family and friends and with strangers, and difficulties in sexual relations; 5) Life activities, referred to the difficulty in the development of housework; and 6) Participation in society, which includes difficulties in participating in community activities, time spent on the disease, emotions, finances, family, and leisure. The Spanish version of the questionnaire by Vázquez-Barquero, Herrera, Vázquez, and Gaité (2006) was used in the present study. The reliability reported by these authors was high (up to .80) for five of the six domains and for the global scales. The "Getting along with people" domain had a value of .76. Internal consistency indicated very high Cronbach  $\alpha$  values for the global scales (.93) and Cronbach values of up to .70 for each of the domains.

The SIP is an instrument that measures behavioral changes related to health and dysfunction. A total of 136 items and 12 categories comprise the instrument. The categories include: 1) Sleep and rest; 2) Eating; 3) Work; 4) Home management; 5) Recreation and pastimes; 6) Ambulation; 7) Mobility; 8) Body care and movement; 9) Social interaction, referring to a reduction in social visits, a reduction in group social activities, decreased sexual activity, and decreased interaction with people; 10) Alertness behavior, related to difficulty in reasoning and problem solving, memory and/or concentration problems, and an increased number of minor accidents; 11) Emotional behavior, which is related to anxiety, complaints of pain or discomfort, and hopelessness about the future; and 12) Communication. A physical domain, a psychosocial domain, and a total index are also included. Scores range from 0–100, with a higher score indicating a worse health status. The Spanish version shows a high internal consistency (.95). Reliability is also high, with a Cronbach alpha of .96 (Badía & Alonso, 1994).

The SF-36 was created by the Medical Outcome Study (MOS). It measures both mental and physical

status through eight dimensions: physical functioning, role-physical, bodily pain, general health, vitality, social functioning, role-emotional, and mental health. Scores range from 0 to 100, with higher scores indicating a better HRQoL. Internal consistency values are up to .70 in all the dimensions, with a range between .71 and .94 (Alonso, Prieto, & Antó, 1995; Ayuso-Mateos, Lasa, Vázquez-Barquero, Oviedo, & Díez-Manrique, 1999).

Finally, a semi-structured interview was created to assess satisfaction with the teleassistance program. Questions about satisfaction with group sessions, topics discussed, support material, and the teleassistance program used were asked.

### Procedure

To conduct the present study, a quasi-experimental static-group comparison design with pre-test and post-test was conducted. Several psychometric tests were administered before and after the online intervention to compare both measures and assess if statistically significant differences appeared. The time between these assessments was approximately 4.5 months.

In a first stage, a web site was created (<http://neuromusculares.deusto.es>). On this web page, participants had access to information about their own disease, a forum, and the videoconference program. Before the program was launched online, an assessment was conducted in person for both groups (experimental and control) by psychologists at the aforementioned hospitals and associations, as well as at the Universidad de Deusto. Participants were assigned to one of two groups by convenience: a videoconference intervention group and a control group, taking into account the availability of a computer and a webcam to be assigned to the first one. Intervention had a total duration of three months. Seven one-hour sessions were conducted, one every two weeks. The intervention was conducted by an expert psychologist via the Skype computer program in group videoconference sessions of four to five people. Sessions were conducted in an experimental room, from which participants were video-called. They remained in their homes participating with their personal computers. The goal of the program was to create a social network between affected people and to improve their HRQoL. The online psychosocial program was based on Cognitive-Behavioral Therapy (CBT), and involved several areas: First, an assessment of the expectations towards the online intervention was carried out in order to identify and clarify the aims of the psychosocial program. Second, a psychoeducation on relevant aspects of neuromuscular diseases was conducted (the explanation of symptoms, changes in the familiar and personal area, exacerbations, or daily obstacles). The third area to

discuss about was emotional reactions to NDs, where the aims were to identify emotional reactions to the disease, to differentiate between healthy and unhealthy emotional expressions, to manage unhealthy emotions, and to strengthen the use of adaptive emotions when communicating. In the fourth area of the intervention a guided imagery to reduce stress and anxiety levels was explained. An audio file was sent by email with instructions to achieve a relaxation status. Fifth, to increase the perception of control participants were said to identify negative automatic thoughts related to the ND, to modify them and to replace them with more adaptive ones (cognitive restructuring, based on Beck cognitive therapy) in order to reduce the feeling of uncertainty associated with disease evolution and future quality of life. Finally, participants were provided with problem solving strategies. Problems related to daily life with the disease or with other areas were classified and tackled with strategies based on the D'Zurilla and Nezu model. In addition, home tasks were put such as self-reports about thoughts, emotional reactions or important problems.

Individuals who did not know how to use Skype or how to navigate the Internet or the study's website received a telephone call to guide them through the program and to solve their doubts. Participants could also communicate with the team by e-mail to get their questions answered. After the online program was completed, a post-test was conducted that utilized the same instruments used in the pre-test stage and a final interview to assess satisfaction with the teleassistance program.

### Data analysis

Statistical analysis was performed using the SPSS v.18 program. Chi square test and Student's t-test were used to verify the homogeneity between the experimental and the control groups before and after the intervention program. A non-parametric Mann-Whitney U test was performed to analyze the inter-group variability, and the Wilcoxon test was also conducted to assess the intra-group variability between the pre-test and post-test scores. *P*-values < .05 were considered to indicate significance. The effect size was also calculated in order to calculate the strength of the correlations obtained.

### Results

The analysis of the data indicated that the experimental and the control group were homogeneous in the following variables assessed: gender ( $\chi^2(1) = .573$ ,  $p = .449$ ), type of ND ( $\chi^2(8) = 9.209$ ,  $p = .325$ ), other non-NDs ( $\chi^2(1) = .025$ ,  $p = .873$ ), marital status ( $\chi^2(5) = 2.152$ ,  $p = .828$ ), employment situation ( $\chi^2(8) = 14.359$ ,

$p = .073$ ), and psychological and/or psychiatric support ( $\chi^2(1) = .402$ ,  $p = .526$ ).

The inter-group variability for the pre-test stage revealed no significant differences between the EG and CG in the following areas: Psychosocial area ("Participation in society", "Alertness behavior", "Emotional behavior", "Recreation and pastimes", "Role Emotional", "Mental Health", "Mental Scale" and "Psychosocial Domain"), Physical area ("Getting around", "Life activities", "Sleep and rest", "Body care and movement", "Home management", "Ambulation", "Communication", "Work", "Eating", "Physical Domain", "Physical Functioning", "Role Physical", "Bodily Pain", "Vitality" and "Physical Scale") and General Health area ("Total WHODAS Working" and "General Health"). Otherwise, significant differences were found ( $p \leq .05$ ) in the social domain, cognitive domain, physical domain and general health domain. In the social domain, higher scores were obtained in the EG compared to the CG in the variables "Getting along with people", "Social Interaction" and "Social Functioning". In the cognitive domain, results indicated higher scores in the EG in "Understanding and communicating". In the physical domain, higher scores were found in the EG in "Self-care" and "Mobility". Finally, the general health domain revealed higher scores in the EG compared to the CG in "Total WHODAS Not Working" and "Total SIP". Medium effect sizes were obtained for the aforementioned variables (see table 1). No significant differences were found for/in the inter-group variability of the post-test stage in all the variables assessed (see Table 2).

The intra-group variability between the pre-test and post-test mean scores indicated significant differences in the following variables of the EG.

Considering the results obtained from the WHO-DAS II questionnaire analysis, it can be concluded that significant differences were found between the pre-test and post-test scores of the experimental group for several domains of this instrument ( $p \leq .05$ ). Specifically, as reported in table 3, the scores for the domains "Understanding and communicating", "Getting along with people", "Life activities", "Participation in society", "Total WHODAS working", and "Total WHODAS not working" were reduced in the post-test stage. A medium effect size ranging between  $r = -.42$  and  $r = -.58$  was obtained for all domains with the exception of "Total WHODAS Working" and "Total WHODAS Not Working" which yielded a larger effect ( $r = -.79$ ,  $r = -.69$  respectively). Significant differences between the pre-test and post-test scores were not found for the control group for most of the WHODAS-II domains except "Life activities", the score of which increased from pre- to post-test ( $z = -1.960$ ,  $r = -.43$ ,  $p \leq .05$ ).

**Table 1.** Comparison between the mean scores of the EG and the CG: Pre-test

Pre-test Variables	Experimental Group	Control Group	U	Z	P-value	r
	Mean ± SD	Mean ± SD				
<i>WHO-DAS II</i>						
Understanding and communicating	14.58 ± 17.37	4.28 ± 8.98	139.00	-2.745	.006**	-.41
Self-care	20.83 ± 24.30	6.66 ± 15.91	175.00	-1.980	.048*	-.30
Getting along with people	17.36 ± 22.10	5.15 ± 11.62	151.00	-2.533	.011*	-.38
Total WHODAS Not Working	34.71 ± 18.96	14.47 ± 12.38	47.50	-3.038	.002**	-.45
<i>SIP</i>						
Mobility	10.41 ± 12.67	1.42 ± 4.78	146.00	-2.945	.003**	-.44
Social interaction	21.87 ± 19.04	9.04 ± 11.68	160.50	-2.137	.033*	-.32
Total SIP	20.68 ± 14.75	11.44 ± 8.40	155.50	-2.198	.028*	-.33
<i>SF-36</i>						
Social Functioning	64.58 ± 27.25	81.54 ± 26.10	152.50	-2.318	.020*	-.35

Notes: EG = Experimental Group; CG = Control Group; SD = Standard Deviation; U = Mann-Whitney U value; r = Effect size.

\* $p \leq .05$ .

\*\* $p \leq .01$ .

Taking into account the SIP results in the experimental group, the scores in five categories of this instrument were significant decreased post-test compared with pre-test ( $p \leq .05$ ). The effect size obtained was medium and ranged from  $-.40$  to  $-.51$ . This significant reduction occurred in "Emotional behavior", "Social interaction", "Alertness behavior", "Psychosocial Domain" and "Total SIP" (see table 3). There were no significant differences in any categories for the control group ( $p > .05$ ).

The SF-36 questionnaire results indicated a statistically significant increase in the post-test score compared with the pre-test score on the "General Health" scale ( $z = -2.138$ ,  $r = -.44$ ,  $p \leq .05$ ) (see table 3).

The control group did not have significantly different pre-test and post-test scores on this scale ( $p > .05$ ).

Regarding satisfaction with the teleassistance program, 84.85% of the participants reported high levels of satisfaction with the group sessions, and 95.85% reported that the topics discussed were adequate. Finally, 83.3% of participants reported that they would participate in future projects involving videoconferencing.

## Discussion

The main aim of the present study was the development of an online tool that would support people with NDs,

**Table 2.** Comparison between the mean scores of the EG and the CG: Post-test

Post-test Variables	Experimental Group	Control Group	U	Z	P-value
	Mean ± SD	Mean ± SD			
<i>WHO-DAS II</i>					
Understanding and communicating	7.50 ± 11.42	5.95 ± 9.69	238.00	-.360	.719
Self-care	17.08 ± 22.16	12.85 ± 21.24	225.00	-.668	.504
Getting along with people	7.98 ± 11.90	7.53 ± 15.34	230.00	-.560	.579
Total WHODAS Not Working	25.00 ± 17.79	16.30 ± 14.55	92.50	-1.341	.180
<i>SIP</i>					
Mobility	9.58 ± 13.01	5.23 ± 12.89	189.50	-1.699	.089
Social interaction	13.33 ± 16.26	6.42 ± 8.23	198.50	-1.274	.203
Total SIP	15.80 ± 12.15	10.04 ± 8.25	190.00	-1.412	.158
<i>SF-36</i>					
Social Functioning	66.66 ± 27.25	80.95 ± 22.57	176.50	-1.759	.079

Notes: EG = Experimental Group; CG = Control Group; SD = Standard Deviation; U = Mann-Whitney U value.

\* $p \leq .05$ .

\*\* $p \leq .01$ .

**Table 3.** Pre-test and post-test mean scores: Experimental group

Variables	EG pre-test		EG post-test		Z	P-value	r
	Mean ± SD	n	Mean ± SD	n			
<i>WHO-DAS II</i>							
Understanding and communicating	14.58 ± 17.37	24	7.50 ± 11.42	24	-2.076	.038*	-.42
Self-care	20.83 ± 24.30	24	17.08 ± 22.16	24	-1.308	.191	-
Getting along with people	17.36 ± 22.10	24	7.98 ± 11.9	24	-2.289	.022*	-.47
Life activities	43.33 ± 37.49	24	34.16 ± 36.82	24	-2.844	.004**	-.58
Participation in society	34.72 ± 26.57	24	26.38 ± 23.84	24	-2.052	.040*	-.42
Total WHODAS Working	13.32 ± 17.47	8	11.20 ± 15.26	8	-2.226	.026*	-.79
Total WHODAS Not Working	34.71 ± 18.96	16	25.00 ± 17.79	16	-2.768	.006**	-.69
<i>SIP</i>							
Emotional behavior	20.83 ± 19.47	24	13.88 ± 18.89	24	-1.974	.048*	-.40
Mobility	10.41 ± 12.67	24	9.58 ± 13.01	24	-.426	.670	-
Social interaction	21.87 ± 19.04	24	13.33 ± 16.26	24	-2.489	.013*	-.51
Alertness behavior	23.33 ± 23.71	24	13.33 ± 14.93	24	-2.289	.022*	-.47
Psychosocial Domain	19.96 ± 16.47	24	12.76 ± 12.10	24	-2.404	.016*	-.49
Total SIP	20.68 ± 14.75	24	15.80 ± 12.15	24	-2.252	.024*	-.46
<i>SF-36</i>							
General Health	37.20 ± 21.40	24	43.75 ± 17.76	24	-2.138	.033*	-.44
Social Functioning	64.58 ± 27.25	24	66.66 ± 27.25	24	-.556	.578	-

Notes: EG = Experimental Group; SD = Standard Deviation; r = Effect size.

\* $p \leq .05$ .

\*\* $p \leq .01$ .

help develop their social networking, and improve their HRQoL. A pre/post comparison of measures on three psychometric instruments was performed to identify any changes in participants' psychosocial health. A control group was included to examine the teleassistance program's effectiveness in a more reliable manner.

Results showed a greater disability and worse HRQoL in the EG before intervention compared to the CG in the following areas: psychosocial, physical and general health. After the teleassistance program, scores in these areas were homogeneous in both groups due to the improvement in the HRQoL of the EG.

In the psychosocial area there was an improvement in concentration, memory, problem solving, social interaction, group social activities, mood and complaints of pain or discomfort. Clinically, when the scores of the variables included in this area are compared to those from other authors with similar pathologies (Leonardi et al., 2010; Vázquez-Barquero et al., 1991), some aspects should be highlight. First, before intervention, participants of the actual study had higher levels of disability than those reported by these authors. Second, after intervention, lower levels of disability were obtained for the present study. In the physical area, a reduction in the difficulties when developing housework properly was obtained. Clinically, after intervention, participants in the actual study had worse disability than that reported by Leonardi et al. (2010), in spite of the

improvement in such area. Finally, the general health area also indicated an improvement in HRQoL levels in the intervention group. Clinically, before intervention, participants' general health was worse or similar in this area than that reported by other authors (Leonardi et al., 2010; Tworck et al., 2010; Vázquez-Barquero et al., 1991; Winter et al., 2010). After intervention, participants of the actual study obtained a similar or better general health compared to the studies cited. The control group did not obtain any relevant significant change in these areas.

These findings suggest that the online program applied is a feasible tool at improving psychosocial, physical and general health aspects, decreasing illness impact and increasing HRQoL of people with rare NDs. These results are congruent with those found by other authors (Amayra et al., 2008; Lázaro et al., 2009; López et al., 2009). The lack of studies analyzing video-conference and HRQoL in people with rare NDs leads us to take into account studies of videoconferencing psychotherapy in non NDs. Shepherd et al. (2006) concluded that a psychological treatment by videoconferencing improved quality of life in people with cancer disease. Emotional and functional well-being were specially benefited after the intervention. Social and physical well-being were indirectly improved.

One of the main aims of the actual study was to create social networks in order to increase social interaction

and social support between people affected with a similar rare ND. Increasing social support can be considered a primary aim when improving HRQoL. Schwartz and Frohner (2005) highlight the significant contribution of this variable to the quality of life, indicating its positive correlation with mental health levels in people with MS. Wilson, Washington, Engel, Ciol, and Jensen (2006) also indicate that perceived social support is a significant variable in the prediction of mental health in people with NDs. In fact, their findings reveal a better psychological adjustment with a higher perceived social support.

The intervention of the actual study is based on CBT, which seems to be an ideal model to improve HRQoL in people with NDs (Cosio, Jin, Siddique, & Mohr, 2011). In this sense, it can be reported the effectiveness of psychological and social intervention programs at improving HRQoL in this population (Hart, Fonareva, Merluzzi, & Mohr, 2005).

In summary, changes after the teleassistance program were significant in the experimental group in psychosocial, physical and general health aspects, indicating a reduction in the subjective level of disability and an improvement in HRQoL. Of note, the results reveal that social functioning is an important variable related to HRQoL and people with NDs (Graham, Rose, Grunfeld, Kyle, & Weinman, 2011).

New technologies seem to be a suitable method of intervention in people with NDs. Indeed, participants' satisfaction with the videoconferencing program was very high and was similar to that found by Doorenbos et al. (2010), Gustke et al. (2000), Mair and Whitten (2000), and Vitacca et al. (2010).

Certain limitations should be considered when interpreting the results of this study. First, sample was not randomized. Second, there were some difficulties obtaining the sample because of diagnosis delay, geographic dispersion, and low disease prevalence. However, the sample size is sufficient, considering the prevalence of NDs in the general population (ASEM/AFM, 2004; Emery, 1991). Third, only participants who had access to a computer and to the Internet were benefited. Fourth, computer-illiterate participants did not benefit; they were included in the control group. Finally, a three, six and twelve months' follow-up would be necessary to assess if these changes remain over time.

In conclusion, the results showed that seven-session videoconferencing effectively improved HRQoL levels in people with NDs. Social interaction appears to play a significant role in it. Further research including the use of teleassistance as a complementary tool for intervention and assessment would be useful in order to enhance the knowledge of NDs and HRQoL.

## References

- Alonso J., Prieto L., & Antó J. M. (1995). La versión española del SF-36 Health Survey (Cuestionario de Salud SF-36): Un instrumento para la medida de los resultados clínicos [The Spanish version of the SF-36 Health Survey: An instrument for measuring clinical outcomes]. *Medicina Clínica*, *104*, 771–776.
- Amayra Caro I., Lázaro Pérez E., López Paz J. F., & De la Cruz Beldarrain A. (2008). On-line psychological support for parents of children and teenagers with neuromuscular diseases. *Neuromuscular Disorders*, *18*, 821. <http://dx.doi.org/10.1016/j.nmd.2008.06.335>
- ASEM (2008). *Guía de las enfermedades neuromusculares. Información y apoyo a las familias* [Neuromuscular diseases guide. Information and support for families]. Jaén, Spain: Formación Alcalá.
- ASEM/AFM (2004). *Enfermedades neuromusculares, 49 fichas* [Neuromuscular diseases, 49 files]. Barcelona, Spain: ASEM. Asociación Española contra las Enfermedades Neuromusculares.
- Ayuso-Mateos J. L., Lasa L., Vázquez-Barquero J. L., Oviedo A., & Díez-Manrique J. F. (1999). Measuring health status in psychiatric community surveys: Internal and external validity of the Spanish version of the SF-36. *Acta Psychiatrica Scandinavica*, *99*, 26–32. <http://dx.doi.org/10.1111/j.1600-0447.1999.tb05381.x>
- Backhaus A., Agha Z., Maglione M. L., Repp A., Ross B., Zuest D., ... Thorp S. R. (2012). Videoconferencing psychotherapy: A systematic review. *Psychological Services*, *9*, 111–131. <http://dx.doi.org/10.1037/a0027924>
- Badía X., & Alonso J. (1994). Adaptación de una medida de la discapacidad relacionada con la enfermedad: La versión española del Sickness Impact Profile [Adapting a measure of dysfunction related to the disease: The Spanish version of the Sickness Impact Profile]. *Medicina Clínica*, *102*, 90–95.
- Bergner M., Bobbitt R. A., Pollard W. E., Martin D. P., & Gilson B. S. (1976). The Sickness Impact Profile: Validation of a health status measure. *Medical Care*, *14*, 57–67. <http://dx.doi.org/10.1097/00005650-197601000-00006>
- Boeschoten R. E., Nieuwenhuis M. M., van Oppen P., Uitdehaag B. M. J., Polman C. H., Collette E. H., ... Dekker J. (2012). Feasibility and outcome of a web-based self-help intervention for depressive symptoms in patients with multiple sclerosis: A pilot study. *Journal of the Neurological Sciences*, *315*, 104–109. <http://dx.doi.org/10.1016/j.jns.2011.11.016>
- Chumbler N. R., Rose D. K., Griffiths P., Quigley P., McGee-Hernandez N., Carlson K. A., ... Hoenig H. (2010). Study protocol: Home-based telehealth stroke care: A randomized trial for veterans. *Trials*, *11*, 74. <http://dx.doi.org/10.1186/1745-6215-11-74>
- Cosio D., Jin L., Siddique J., & Mohr D. C. (2011). The effect of telephone-administered cognitive-behavioral therapy on quality of life among patients with multiple sclerosis. *Annals of Behavioral Medicine*, *41*, 227–234. <http://dx.doi.org/10.1007/s12160-010-9236-y>
- Doorenbos A. Z., Eaton L. H., Haozous E., Towle C., Revels L., & Buchwald D. (2010). Satisfaction with telehealth for cancer support groups in rural American Indian and Alaska

- native communities. *Clinical Journal of Oncology Nursing*, 14, 765–770. <http://dx.doi.org/10.1188/10.CJON.765-770>
- Emery A.** (1991). Population frequencies of inherited neuromuscular diseases—A world survey. *Neuromuscular Disorders*, 1, 19–29. [http://dx.doi.org/10.1016/0960-8966\(91\)90039-U](http://dx.doi.org/10.1016/0960-8966(91)90039-U)
- Ferrer-Roca O., García-Nogales A., & Peláez C.** (2010). The impact of telemedicine on quality of life in rural areas: The Extremadura model of specialized care delivery. *Telemedicine and e-Health*, 16, 233–243. <http://dx.doi.org/10.1089/tmj.2009.0107>
- Fioravanti G., Casale S., Mantegazza R., Leonardi M., & Raggi A.** (2010). Self-efficacy, social support and locus of control as correlates of health-related quality of life in myasthenia gravis. *Bollettino di Psicologia Applicata*, 261, 19–27.
- Graham C. D., Rose M. R., Grunfeld E. A., Kyle S. D., & Weinman J.** (2011). A systematic review of quality of life in adults with muscle disease. *Journal of Neurology*, 258, 1581–1592. <http://dx.doi.org/10.1007/s00415-011-6062-5>
- Gustke S. S., Balch D. C., West V. L., & Rogers L. O.** (2000). Patient satisfaction with telemedicine. *Telemedicine Journal*, 6, 5–13. <http://dx.doi.org/10.1089/107830200311806>
- Hart S., Fonareva I., Merluzzi N., & Mohr D. C.** (2005). Treatment for depression and its relationship to improvement in quality of life and psychological well-being in multiple sclerosis patients. *Quality of Life Research*, 14, 695–703. <http://dx.doi.org/10.1007/s11136-004-1364-z>
- Hubbard P. A., Broome M. E., & Antia L. A.** (2005). Pain, coping, and disability in adolescents and young adults with cystic fibrosis: A Web-based study. *Pediatric Nursing*, 31, 82–86.
- Kierkegaard M., Harms-Ringdahl K., Holmqvist L. W., & Tollbäck A.** (2011). Functioning and disability in adults with myotonic dystrophy type 1. *Disability and Rehabilitation*, 33, 1826–1836. <http://dx.doi.org/10.3109/09638288.2010.549287>
- Kroenke K., Theobald D., Wu J., Norton K., Morrison G., Carpenter J., & Tu W.** (2010). Effect of telecare management on pain and depression in patients with cancer: A randomized trial. *JAMA: Journal of the American Medical Association*, 304, 163–171. <http://dx.doi.org/10.1001/jama.2010.944>
- Lázaro Pérez E., Amayra Caro I., López Paz J. F., De la Cruz Beldarrain A., & Alday E.** (2009). Psychosocial online program directed to parents of children and teenagers with neuromuscular disease. *Journal of Health Technology and Application*, 7, 78–81.
- Leonardi M., Raggi A., Antozzi C., Confalonieri P., Maggi L., Cornelio F., & Mantegazza R.** (2009). Identification of international classification of functioning, disability and health relevant categories to describe functioning and disability of patients with myasthenia gravis. *Disability and Rehabilitation*, 31, 2041–2046. <http://dx.doi.org/10.1080/09638280902893634>
- Leonardi M., Raggi A., Antozzi C., Confalonieri P., Maggi L., Cornelio F., & Mantegazza R.** (2010). The relationship between health, disability and quality of life in myasthenia gravis: Results from an Italian study. *Journal of Neurology*, 257, 98–102. <http://dx.doi.org/10.1007/s00415-009-5279-z>
- López Paz J. F., Amayra Caro I., Lázaro Pérez E., De la Cruz Beldarrain A., & Alday E.** (2009). Effectiveness of a psycho-educational online program directed to children and teenagers with neuromuscular disease. *Journal of eHealth Technology and Application*, 7, 72–77.
- Macea D. D., Gajos K., Daglia Calil Y. A., & Fregni F.** (2010). The efficacy of Web-based cognitive behavioral interventions for chronic pain: A systematic review and meta-analysis. *The Journal of Pain*, 11, 917–929. <http://dx.doi.org/10.1016/j.jpain.2010.06.005>
- Mair F., & Whitten P.** (2000). Systematic review of studies of patient satisfaction with telemedicine. *BMJ*, 320, 1517–1520. <http://dx.doi.org/10.1136/bmj.320.7248.1517>
- Polisena J., Tran K., Cimon K., Hutton B., McGill S., & Palmer K.** (2009). Home telehealth for diabetes management: A systematic review and meta-analysis. *Diabetes, Obesity & Metabolism*, 11, 913–930. <http://dx.doi.org/10.1111/j.1463-1326.2009.01057.x>
- Polisena J., Tran K., Cimon K., Hutton B., McGill S., Palmer K., & Scott R. E.** (2010). Home telehealth for chronic obstructive pulmonary disease: A systematic review and meta-analysis. *Journal of Telemedicine & Telecare*, 16, 120–127. <http://dx.doi.org/10.1258/jtt.2009.090812>
- Rabinowitz T., Murphy K. M., Amour J. L., Ricci M. A., Caputo M. P., & Newhouse P. A.** (2010). Benefits of a telepsychiatry consultation service for rural nursing home residents. *Telemedicine and e-Health*, 16, 34–40. <http://dx.doi.org/10.1089/tmj.2009.0088>
- Rotondi A. J., Sinkule J., & Spring M.** (2005). An interactive web-based intervention for persons with TBI and their families: Use and evaluation by female significant others. *Journal of Head Trauma Rehabilitation*, 20, 173–185. <http://dx.doi.org/10.1097/00001199-200503000-00005>
- Schwartz C., & Frohner R.** (2005). Contribution of demographic, medical, and social support variables in predicting the mental health dimension of quality of life among people with multiple sclerosis. *Health & Social Work*, 30, 203–212. <http://dx.doi.org/10.1093/hsw/30.3.203>
- Shepherd L., Goldstein D., Whitford H., Thewes B., Brummell V., & Hicks M.** (2006). The utility of videoconferencing to provide innovative delivery of psychological treatment for rural cancer patients: Results of a pilot study. *Journal of Pain and Symptom Management*, 32, 453–461. <http://dx.doi.org/10.1016/j.jpainsymman.2006.05.018>
- Sloan D. M., Gallagher M. W., Feinstein B. A., Lee D. J., & Pruneau G. M.** (2011). Efficacy of telehealth treatments for posttraumatic stress-related symptoms: A meta-analysis. *Cognitive Behaviour Therapy*, 40, 111–125. <http://dx.doi.org/10.1080/16506073.2010.550058>
- Soutter J., Hamilton N., Russell P., Russell C., Bushby K., Sloper P., & Bartlett K.** (2004). The Golden Freeway: A preliminary evaluation of a pilot study advancing information technology as a social intervention for boys with Duchenne muscular dystrophy and their families. *Health and Social Care in the Community*, 12, 25–33. <http://dx.doi.org/10.1111/j.1365-2524.2004.00465.x>



- Twork S., Wiesmeth S., Klewer J., Pöhlau D., & Kugler J.** (2010). Quality of life and life circumstances in German myasthenia gravis patients. *Health and Quality of Life Outcomes*, 8, 129. <http://dx.doi.org/10.1186/1477-7525-8-129>
- Vázquez-Barquero J. L., Arias M. A., Peña C., Díez J. F., Ayesterán A., & Miró J.** (1991). El cuestionario “perfil de impacto de la enfermedad” (SIP): Versión española de una medida del estado de salud [The Sickness Impact Profile questionnaire (SIP): Spanish versión of a health status measure]. *Actas Luso-Españolas de Neurología, Psiquiatría y Ciencias Afines*, 19, 127–134.
- Vázquez-Barquero J. L., Herrera S., Vázquez E., & Gaité L.** (2006). *Cuestionario para la evaluación de discapacidad de la Organización Mundial de la Salud WHO-DAS II* [World Health Organization disability assessment schedule WHO-DAS II]. Madrid, Spain: Subdirección General de Información Administrativa y Publicaciones.
- Vitacca M., Bianchi L., Guerra A., Fracchia C., Spanevello A., Balbi B., & Scalvini S.** (2009). Tele-assistance in chronic respiratory failure patients: A randomized clinical trial. *European Respiratory Journal*, 33, 411–418. <http://dx.doi.org/10.1183/09031936.00005608>
- Vitacca M., Comini L., Tentorio M., Assoni G., Trainini D., Fiorenza D., ... Scalvini S.** (2010). A pilot trial of telemedicine-assisted, integrated care for patients with advanced amyotrophic lateral sclerosis and their caregivers. *Journal of Telemedicine and Telecare*, 16, 83–88. <http://dx.doi.org/10.1258/jtt.2009.090604>
- Ware J. E., & Sherbourne C. D.** (1992). The MOS 36-item short-form health-survey (SF-36): I. Conceptual framework and item selection. *Medical Care*, 30, 473–483. <http://dx.doi.org/10.1097/00005650-199206000-00002>
- Watson A., Bickmore T., Cange A., Kulshreshtha A., & Kvedar J.** (2012). An Internet-based virtual coach to promote physical activity adherence in overweight adults: Randomized controlled trial. *Journal of Medical Internet Research*, 14, 44–55. <http://dx.doi.org/10.2196/jmir.1629>
- Wilson S., Washington L. A., Engel J. M., Ciol M. A., & Jensen M. P.** (2006). Perceived social support, psychological adjustment, and functional ability in youths with physical disabilities. *Rehabilitation Psychology*, 51, 322–330. <http://dx.doi.org/10.1037/0090-5550.51.4.322>
- Winter Y., Schepelmann K., Spottke A. E., Claus D., Grothe C., Schröder R., & Dodel R.** (2010). Health-related quality of life in ALS, myasthenia gravis and facioscapulohumeral muscular dystrophy. *Journal of Neurology*, 257, 1473–1481. <http://dx.doi.org/10.1007/s00415-010-5549-9>
- World Health Organization** (2001). *World Health Organization Disability Assessment Schedule II (WHODAS II)*. Geneva, Switzerland: World Health Organization.