

ORIGINAL ARTICLE

Integrated care of muscular dystrophies in Italy. Part 2. Psychological treatments, social and welfare support, and financial costs

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This paper describes the psycho-social treatments received by 502 patients with MDs and their relatives, and the costs for care sustained by the families in the previous six month period.

Data were collected by the MD-Care Schedule (MD-CS) and the Family Problems Questionnaire (FPQ).

Psycho-educational interventions were provided to 72 patients (14.3%), and social/welfare support to 331 patients (65.9%). Social/welfare support was higher in patients with DMD or LGMD, in those showing more severe disability, and in patients who were in contact with centres located in Northern Italy.

Psycho-educational interventions were received by 156 (31%) relatives, and social/welfare support by 55 (10.9%) and mainly provided by Family/Patients Associations (83.6%).

Relatives with higher educational levels, who spent more daily hours in the assistance of patients with DMD, and in contact with centres in Central Italy more frequently benefited from psycho-educational interventions.

In the previous year, costs for care were sustained by 314 (63.9%) relatives. Financial difficulties related to patient's condition, were higher in families of patients who needed more intensive rehabilitation and daily hours of caregiving,

and in families who lived further away from the reference's centre.

These results showed that psycho-social aspects of MDs care are only partially met in Italy, and that ad hoc supportive interventions for these patients and their families should be potentiated.

Key words: muscular dystrophies, psycho-social treatments, costs for care

Introduction

We have recently shown that the clinical management of MDs in Italy is complex and varies in relation to functional disabilities and type of MD, pattern of care and geographical area (1).

This paper focuses on the psycho-social management of MDs and on the financial costs for care sustained by the families.

Patients and methods

Details on participating centers, study design, and assessment instruments are reported by Magliano et al. (2).

The sample consisted of 502 key-relatives, of which the majority were mothers (84.6%) and cohabiting (87.8%) and had a mean age of 43.4 (7.4) years. Almost half of them had received higher education (56.3%) and were employed (52.6%). They spent on average 5.7 (4.6sd) daily hours in patient's care-giving in the previous two months. Of the 502 patients, most of them were male (96.4%), and school attending (85.6%). Three-hundred-thirty-three (66.3%) of them suffered from DMD, 129 (25.7%) from BMD, and 40 (8.0%) from LGMDs. The mean level of independence in daily activities, measured by the BI, was 68.3 (31.3sd). One-hundred-ninety-four patients (38.6%) were in wheelchair. Three-hundred-sixty-nine (73.5%) patients assumed drugs, and 351 (69.9%) attended rehabilitation treatment. Patients lived, on average, 183.9 (255.7 sd) kilometres far away from the reference centre.

Statistical analysis

Differences in psychosocial treatments, social/welfare support and financial difficulties in relation to patients' socio-demographic, clinical, and geographic variables were explored by analysis of variance and χ^2 , as appropriate. Correlations between the above-mentioned variables and patient' socio-demographic and clinical variables were explored by Spearman's r coefficient.

Multiple regression analyses were performed to explore the simultaneous effects on psychosocial treatments, social/welfare support, and financial difficulties (dependent variables) of patients' socio-demographic characteristics and clinical variables (first block), and geographic variables and relatives' perception of support received by social network and professionals (second block). Only variables statistically significant related to the dependent variables in univariate analyses were included in the multivariate ones. Statistical significance was set at $p < 0.01$.

Results

Psycho-educational interventions

Seventy-two patients (14.3%) received a psycho-educational intervention in the six months preceding the interview. Of these, 38 (52.8%) received a psychological support and 28 (38.9%) information on MD treatments.

Of the 156 relatives receiving psycho-educational interventions, 107 (68.5%) were informed on clinical and rehabilitative procedures and 84 (53.8%) on MD treatments; 34 relatives (21.7%) received psychological support (Table 1).

Psycho-educational interventions were more often provided to relatives who spent more daily hours in care-giving ($r = .28$, $p < .0001$), and to relatives of patients younger ($r = -.15$, $p < .001$), with a shorter duration of illness ($r = -.21$, $p < .0001$), and suffering from DMD

Table 1. Psycho-educational interventions, and social/welfare support received by patients with md and their relatives in the past six months (n = 502).

	Patients	Relatives
Psycho-educational intervention Yes, N (%)	72 (14.3)	156 (31.1)
Type of intervention N (%)		
Information on patient's illness	24 (33.4)	64 (41.0)
Information on MD treatments	28 (38.9)	84 (53.8)
Education on clinical and rehabilitative procedures	-	107 (68.5)
Psychological support	38 (52.8)	34 (21.7)
Genetic counselling	10 (13.9)	22 (14.1)
Mean (SD)	0.2 (0.5)	0.6 (1.1)
Social/welfare support Yes, N (%)	331 (65.9)	55 (10.9)
Type of support N (%)		
School support	52 (15.7)	
Health volunteers' help	15 (4.5)	-
Self-help groups	-	12 (21.8)
Family and users associations help	-	46 (83.6)
School transport	51 (15.4)	-
Medical care and rehabilitation transports	21 (6.3)	-
Economic benefits	319 (96.4)	-
Mean (SD)	0.9 (0.9)	0.1 (0.3)

(0.8 (1.2) vs. 0.3 (0.7) vs. 0.2 (0.7), $F = 14.8$, $df 2$, 499, $p < .0001$). Psycho-educational interventions were more frequently available at centres located in central Italy [0.4 (0.8) vs. 1.2 (1.3) vs. 0.3 (0.9), $F = 42.8$, $df 2$, 499, $p < .0001$; 0.3 (0.7) vs. 1.4 (1.3) vs. 0.2 (0.6), $F = 79.6$, $df 2$, 499, $p < .0001$].

Social/welfare support

In the previous six months, 331 patients received social/welfare support, mainly consisting (96.4%) in economic benefits. Other supports, such as school support, were provided to 15.7% of patients only (Table 1).

Patients who were older ($r = .18$, $p < .0001$), had a poorer functional autonomy ($r = -.51$, $p < .0001$), a longer duration of illness ($r = .24$, $p < .0001$), and/or suffering from DMD received more social/welfare support (DMD 1.1 (0.9), BMD 0.4 (0.6), LGMD 0.7 (0.8), $F = 41.7$, $df 2$, 499, $p < .0001$). Furthermore, patients who were in contact with centres located in Northern Italy received more social/welfare support (North 1.2 (1.0), Central 0.7 (0.6), South 0.8 (0.8), $F = 14.9$, $df 2,499$, $p < .0001$).

Only 55 (10.9%) relatives received social/welfare support in the previous six month, mainly provided (83.6%) by Family/Patients Associations. Furthermore, the support was more often provided to relatives of patients who had lower functional autonomy ($r = -.14$, $p < .01$) and/or suffered from DMD or LGMD (DMD 0.1 (0.4), BMD 0.03 (0.2), LGMD 0.1 (0.3), $F = 6.5$, $df 2$, 499, $p < .01$).

Costs and economic difficulties

Three-hundred-fourteen (63.9%) out of 406 relatives reported costs for patient's care in the previous year. Of these, 207 (65.9%) listed costs for medical/nursing care and 201 (64.0%) for drugs. Moreover, 88 (28.1%) relatives reported "other costs" as transfer (36.4%), rehabilitation (26.1%), medical devices (25%), adaptive devices (15.9%), and psychological support (6.8%).

Costs for care ($N = 53.5\%$, $C = 54.0\%$, $S = 75.8\%$, $\chi^2 = 26.6$, $df 2$, $p < .001$; $N = 59.2\%$, $C = 56.7\%$, $S = 75.3\%$, $\chi^2 = 14.7$, $df 2$, $p < .001$), drugs ($N 26.7\%$, $C 34.6\%$, $S 54.4\%$, $\chi^2 = 29.0$, $df 2$, $p < .0001$; and $N 31.2\%$, $C 30.1\%$, $S 55.9\%$, $\chi^2 = 31.8$, $df 2$, $p < .0001$, respectively), doctors and nurses ($N 30.4\%$, $C 35.0\%$, $S 58.6\%$, $\chi^2 = 30.6$, $df 2$, $p < .0001$) $N 39.4\%$, $C 31.3\%$, $S 55.9\%$, $\chi^2 = 21.5$, $df 2$, $p < .0001$, respectively) resulted higher in families of patients living in Southern Italy.

Difficulties related to patient's condition were reported as moderate by 24.7% of relatives and marked by 9.6% in the previous 12 months. These difficulties were higher among relatives of patients who were older, had a longer duration of illness ($r = .18$, $p < .0001$), less au-

tonomy ($r = -.32$, $p < .0001$), requiring more drug therapies ($r = .27$, $p < .0001$) and/or rehabilitative interventions ($r = .32$, $p < .0001$), and in DMD group (1.6 (0.8) vs. 1.2 (0.5), vs. 1.3 (0.6), $F = 15.3$, $df 2$, 497, $p < .0001$). Finally, economic difficulties were higher among relatives who were unemployed (1.6 (0.8) vs. 1.4 (0.7), $df 1$, 498, $p < .01$); who dedicated more daily hours to the patient's assistance ($r = .13$, $p < .01$), had lower level of support by social network ($r = -.16$, $p < .0001$) and professionals ($r = -.14$, $p < .01$) and living further away from the clinical centre of reference ($r = .12$, $p < .01$).

Multivariate analyses

Socio-demographic and clinical variables (block 1) explained 15% of variance in family psycho-educational intervention, while geographical areas and relative's perceived support explained a further 16% of variance (Table 2). In particular, psycho-educational interventions were more frequently reported by relatives of DMD patients, with higher educational levels, who spent more daily hours in the patient daily assistance or lived in central Italy. Thirty-two percent of variance in patient social welfare support was explained by level of disability, type of disease and geographical location. Family financial difficulties were higher in relation to the daily hours for care, poor social network support and longer distance from the reference centre.

Discussion

The results of this study suggest that psycho-social management of MDs is scarcely available in Italy. In particular, only 14% of patients and 31% of relatives received psycho-educational interventions in the reference period. This condition is likely due to the low integration of medical, psychological and social expertise in services for rare diseases (5-8), in line with the poor psychological support available for patients with other severe diseases (3, 4).

The low percentage of patients receiving information could be related to several factors, such as: a) the duration of illness, whereas information is mainly provided in the phase of communication of the diagnosis; b) the fact that most patients were adolescent, an age in which health issues are managed throughout parents (9). The poor availability of professionals trained in the psychological care of children with rare diseases, should explain the low percentage of patients (38/502) receiving such a support (5, 10).

In our sample, family interventions mostly consisted in education on medical procedures and treatments for MDs, while psychological support was provided to 34 caregivers out of 502, only. Providing the relatives with information on MDs and their treatments may reinforce

Table 2. Multiple hierarchical analyses: effects of socio-demographic, clinical variables, and geographic variables on psycho-educational interventions received by the families, social/welfare support provided to patients with MD, and relatives' economic problems due to patient' health care.

	Family psycho-educational interventions		Patient social/welfare support		Family financial difficulties	
	1 st step	2 nd step	1 st step	2 nd step	1 st step	2 nd step
Blocks	Standardized Beta					
1 - Socio-demographic and clinical variables						
Patient's age	-.07	-.15	-.28	-.23	.10	.11
Barthel Index	-	-	-.45 ^d	-.40 ^d	-.16 ^a	-.08
Duration of illness	-.24 ^c	-.12	.15	.12	-.13	-.14
Type of MD- BMD	-.18 ^c	-.15 ^b	-.14 ^a	-.15 ^b	-.06	-.04
Type of MD-LGMD	-.12 ^b	-.15 ^b	-.08	-.07	-.04	-.02
Patient's education	.18	.24	-.19	-.16	-	-
Relative's education	.13 ^c	.10 ^a	-	-	-	-
Daily hours in caregiving	.19 ^d	.10 ^a	-.06	-.01	.11 ^a	.11 ^a
Relative's employment	-	-	-	-	-.05	-.01
Number of drugs	-	-	-	-	-.08	-.03
Number of rehabilitative interventions	-	-	-	-	.13 ^a	-.07
2 - Geographical area and relatives' perceived support						
Centre located in Northern Italy		-.14		.17 ^b		-
Centre located in Central Italy		.35 ^d		-		-
Centre located in Southern Italy		-		.05		-
Family living area: North Italy		.09		-		-
Family living area: South Italy		-.11		-		-
Professional support		-		-		.02
Social network support		-		-		-.26 ^d
Distance from the Centre						.22 ^d
Model's F, df, p<	15.6; 11,380; .0001		20.6; 9, 382; p < .0001		10.0; 12,445; .0001	
R²	.15	.31	.30	.32	.11	.21

^a = p < .05; ^b = p < .01; ^c = p < .001; ^d = p < .0001

their abilities to deal with practical aspects of daily assistance (8, 9). However, medical education is unable to address relatives' needs for psychological support, which remains largely unmet (5, 8). This is particularly worrying, given the high level of psychological burden among the caregivers of patients with MDs (2).

Our data reveal that psycho-educational interventions were more frequently provided to relatives of patients with DMD, and to relatives of patients young and with short duration of illness. Psycho-educational support was found more available for families who lived or were in contact with centres located in Central Italy. This finding is probably related to the greater presence of a Family Association in this geographical area, and a closer collaboration with the NMD Centres (16).

Undoubtedly, the psychological support is crucial in the early phase of these disorders (9, 10), when the diagnosis of

DMD can be associated with feelings of despair and deep worries for the future (10). However, the psychological support is equally important in the subsequent phases of patient' and family adaptation to the disease (10, 11), when clinical conditions of the patient get worse (10, 12) and daily assistance become more demanding for caregivers (2, 5, 6).

As far as social/welfare support to patients, we found that it mainly consisted in economic benefits, while other support such as school support was available only for a minority of patients. The impossibility for the patient to attend school is a critical event for him/her and his/her relatives, both psychologically and practically (11, 12, 14, 17). Therefore, it is important to provide patients with supports facilitating school attendance as longer as possible. These supports should include dedicated school transfer as well as dedicated helper at school, both available for only 15% in our sample.

The possibility to receive more social/welfare supports by patients who live in Northern Italy reflects the differences in health care policies at regional level (18). In fact, in Southern Italy, the health care provision is poorer due to cutting investment in health services, and regional financial deficit (19). This situation leads to inequalities in accessibility and availability of public health services, as supported by the lower levels of perceived professional support among relatives living in this geographical area ($N = 3.2$ (0.6), $C = 3.1$ (0.7), $S = 2.9$ (0.7), $F = 7.6$, $df 2$, 497 , $p < .0001$).

This situation may also explain why about 25% of patients living in Southern Italy are followed by extra-regional centres. Furthermore, inequalities in the Italian health services resources justify the higher costs sustained by families in Southern Italy (19).

Despite its strengths, the study has some methodological limitations, such as the cross-sectional design, and the cases age limited to 4-25 years.

The results of the present study represent a first step to define the needs for care for patients and relatives, and to develop appropriate psycho-social interventions.

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