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Multidisciplinary care use in neurodegenerative complex diseases: The example of progressive supranuclear palsy and advanced Parkinson's disease in real-life

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ABSTRACT

Background: In spite of being considered the gold-standard of care, little is known about the real-life use of in-home and multidisciplinary care in atypical parkinsonism.
Objective: Primary: Examine real-life multidisciplinary care use for Progressive Supranuclear Palsy (PSP). Secondary: a) Compare PSP care to advanced Parkinson's disease (APD) care; (b) Explore demographic and clinical variables associated with care needs in both groups.
Methods: A cross-sectional multicenter observational study enrolled 129 PSP patients and 65 APD patients (Hoehn and Yahr $\geq$3), matched for sex and age. Univariate and multivariate regression analysis were performed.
Results: Over the previous year, 40 % of PSP patients did not encounter a physical therapist, while only one-third met a speech and language therapist and 5 % an occupational therapist. More than 20 % received in-home care and 32 % needed home structural changes. Compared to APD, PSP patients required more day-time, night-time and home structural changes. When considering both PSP and APD in multivariate analysis, reduced functional autonomy and living without a family caregiver were both related to day-time home assistance and to the need of at least one home care service. A PSP diagnosis compared to APD was a risk factor for having at least four multidisciplinary visits in a year. Finally, PSP diagnosis and being from the Northern Italy were significantly related with home structural changes.
Conclusions: There's a significant gap in providing multidisciplinary care for PSP patients. Our findings emphasize the need for a shared, integrated care plan at a national level for patients with atypical parkinsonism.

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1. Introduction

An integrated care based on a multidisciplinary approach, including the coordinated actions of multiple members of a health care team and patients' participation, associated to pharmacological and non-pharmacological interventions, is currently considered the gold standard of care for complex neurodegenerative diseases [1–3]. Patients with advanced Parkinson's disease (APD) and atypical parkinsonism often share common complex care needs [4,5]. However, due to their low prevalence if compared to PD (8.8–10.8/100,000 for progressive supranuclear palsy -PSP-, 3/100,000 for multiple system atrophy and about 120–130/100,000 for PD in Europe), little is known about the real-life use of multidisciplinary care in atypical parkinsonism [6–8].

PSP is a rapidly progressive neurodegenerative disease characterized by postural instability, vertical supranuclear palsy, akinesia and cognitive dysfunction [9]. Herein, we present the results of a sub-study of the Italian PSP Network (PSP-NET) supported by the "Fondazione LIMPE per il Parkinson Onlus" involving 18 Italian movement disorder centers aimed at collecting demographic and clinical information in an ongoing observational longitudinal study.

Specific aims of the present sub-study were to (1) describe the multidisciplinary care approach adopted in real-life setting for PSP patients, including in-home-care services (primary objective); (2) understand the relationship between multidisciplinary and home-care needs with clinical features and disease milestones in PSP patients; (3) compare PSP multidisciplinary care to the APD one, considered as a control group; (4) investigate risks factors for home-care needs in PSP and APD. Our results could help in identifying the gaps to reach a satisfactory level of multidisciplinary care for PSP patients, used as an example of an orphan and complex neurodegenerative disease.

2. Patients and methods

2.1. Study protocol and patient criteria

This is a cross-sectional sub-study conducted in 18 third level movement disorder centers participating in the PSP-NET coordinated by the Center for Neurodegenerative Diseases (CEMAND), University of Salerno, Italy (data downloaded on May 18, 2023). PSP patients were consecutively enrolled and included since December 2020 if (1) they provided written and signed informed consent; (2) were diagnosed with either possible or probable PSP according to the Movement Disorder Society criteria [9]; (3) were accompanied by a caregiver.

APD patients have been enrolled from three selected centers from the PSP-NET, located in the North, Center and South of the country, i.e. Turin, Rome and Salerno, respectively. Inclusion criteria for the PD group were (1) written and signed informed consent; (2) a diagnosis of PD and Hoehn and Yahr (HY) laying between 3 and 5 in the Med ON condition [10,11]. Recruitment ratio was APD:PSP = 1:2. Sex and age at inclusion of the two groups were matched.

The project was approved by the Ethical committee of the coordinating center (number 178 December the 4th, 2020) and, thereafter, by the ones of all participating centers.

2.2. Data collection

All patients have been evaluated by movement disorders experts and underwent the same structured interview prepared by the study investigators (MF and MP) assessing: a) demographic, clinician and treatment data including levodopa equivalent daily dose (LEDD) [12]; b) the presence of in-home care services including day-time and night-time assistance, nurses interventions, meal delivering and structural home adaptations; c) the access to any multidisciplinary care intervention, including the visits done by physicians and health-care professionals, over the previous 12 months; d) if those visits have been made within the National Health System (NHS) or within a private healthcare assistance. The list of investigated multidisciplinary care visits was chosen based on literature recommendations for PD patients and included the intervention of: neurologist, general practitioner (GP), geriatrician, physical and rehabilitation physician, physiotherapist, speech and language therapist (SLT), occupation therapist, psychologist, psychiatrist, botulinum toxin treatment (BNT), social assistance, surgeon, dietician, pulmonologist, gastroenterologist, ophthalmologist, urologist and spiritual assistance [2,13].

Furthermore, all the patients have been evaluated with the 12 item Neuropsychiatric Inventory test (NPI-12) for neuropsychiatric symptoms and the Schwab an England (S&E) scale to assess independency in activity of daily living [14,15]. Motor symptoms have been assessed by means of the PSP rating scale (PSPRS) for PSP and by the Movement Disorder Society-Unified Parkinson's Disease Rating Scale (MDS-UPDRS) part III for APD [16,17]. Based on the PSPRS the following items have been considered to define the presence of clinically significant milestones denoting greater disease severity in PSP patients: item 26 > 2 for gait impairment, items 3 or 13 > 2 for dysphagia, items 8 or 9 > 2 for cognitive impairment and item 12 > 2 for dysarthria [16]. PSP variants have been classified in agreement with the MDS criteria [9,18]. Patients not qualifying for PSP Richardson's syndrome (PSP-RS) phenotype have been grouped into the other variant syndromes of PSP (vPSP) [18].

2.3. Statistical analysis

Descriptive statistics of demographic, clinical and therapeutic data were provided for continuous [mean and standard deviation (SD)] and categorical (count and percentage) variables. Group comparisons were performed by Chi-square test for categorical data and Mann-Whitney U for continuous data. Univariate analysis with day-time assistance (yes/no), at least one in-home care service (yes/no), more than 4 multidisciplinary visits/year (yes/no) and home structural changes (yes/no) as dependent variables and demographic/clinical/social features (sex, age, age at onset, disease duration, living with a caregiver, NPI-12 total score, S&E, PSP diagnosis/variants, PSPRS total score, disease milestones and center) as independent variables were performed for both the PSP group alone and all the patients together (PSP and APD). Subsequently, all the significant factors ($p \leq 0.05$) were included in logistic regression analysis (one for each dependent variable) to calculate OR (95 % CI) adjusted for all possible confounding effects. Multicollinearity analysis was used to check for correlations between variables before performing multiple regression analysis. P value < 0.05 was considered significant. The software SPSS 27.0 (SPSS, Chicago, IL) was used.

3. Results

3.1. Description of the enrolled cohorts

One-hundred and twenty-nine PSP patients (34.1 % women) were enrolled; mean (SD) age and disease duration were 71.6 (6.5) and 2.3 (1.9) years, respectively (Table 1). About two third of the patients had PSP-RS phenotype (78 %). Mean (SD) PSPRS total score was 42.5 (17.3) (Table 1).

Sixty-five APD patients (53.8 % women) were enrolled with longer disease duration if compared to PSP patients ($p < 0.001$). PD patients mean (SD) MDS-UPDRS-III score was 47.5 (11.8) (Table 1).

PSP patients had a significantly lower NPI-12 total score if compared to APD ($p = 0.002$) mainly due to lower severity of hallucinations, depression, anxiety and sleep and night-time behaviour disorders. Likewise, they had a better S&E ($p = 0.024$) and, as expected, a significantly lower LEDD ($p < 0.001$) (Table 1).

Within each group, a low number of patients (about 7 %) was institutionalized and only one patient had edited living will. In terms of caregiving, compared to APD, PSP were less likely assisted by a relative and showed a tendency towards significance for greater mean daily

Table 1
Demographic, clinical and therapeutic data of PSP and APD.

Patients' data	PSP N = 129	APD N = 65	p
Age, yrs	71.6 (6.5)	73.2 (8.0)	0.100
Women, n (%)	44 (34.1)	35 (53.8)	0.250
Age at onset, yrs	69.6 (6.9)	61.3 (10.3)	**<0.001**
Disease duration, yrs	2.3 (1.9)	11.8 (6.6)	**<0.001**
LEDD	403 (484)	857 (383)	**<0.001**
S&E	53.0 (26.1)	43.0 (21.7)	**0.024**
MDS-UPDRS part III	NA	47.5 (11.8)	/
PSP-rs	42.5 (17.3)	NA	/
NPI-12			
Total score	16.5 (15.8)	24.7 (18.8)	**0.002**
Delusions	0.3 (1.3)	0.6 (2.1)	0.250
Hallucinations	0.1 (0.7)	1.2 (2.3)	**<0.001**
Agitation/Aggression	1.0 (2.1)	1.4 (2.7)	0.360
Depression	2.1 (2.9)	4.1 (4.2)	**<0.001**
Anxiety	1.6 (2.6)	3.1 (4.0)	**0.010**
Elation/Euphoria	0.3 (1.3)	0.7 (0.4)	0.220
Apathy/indifference	3.5 (3.5)	2.5 (4.0)	0.100
Disinhibition	0.4 (1.1)	0.3 (1.1)	0.150
Irritability/Lability	1.3 (2.3)	1.5 (2.8)	0.500
Motor aberrant behaviour	0.8 (2.0)	1.1 (2.6)	0.300
Sleep and Night-time Behaviour Disorders	1.2 (2.5)	2.9 (4.0)	**0.003**
Appetite and Eating Disorders	0.5 (1.1)	0.7 (1.4)	0.320
Institutionalized, n (%)	9 (6.9)	4 (6.1)	0.640
Caregiver			
Hours/day, n	17.9 (8.5)	15.7 (7.9)	0.080
Relative, n (%)	95 (73.6)[a]	61 (94)	**0.005**
Other, n (%)	13 (10)[a]	4 (6.1)	0.361
Living will	1 (0.7)	1 (1.5)	0.900

Values are presented as mean (SD) if no otherwise specified.
Legend: APD: advanced Parkinson's disease; HY: Hoehn Yahr Stage; LEDD: levodopa equivalent daily dose; MDS-UPDRS part III: the Movement Disorder Society sponsored version of the Unified Parkinson's disease rating scale part III; NPI-12: Neuropsychiatric Inventory test 12-items; PSP: Progressive Supranuclear Palsy; PSP-rs: Progressive Supranuclear Palsy rating scale; RS: Richardson's syndrome; S&E: Schwab and England score; vPSP: the other variant syndromes of PSP.
[a] Missing data for 21 PSP

Table 2
In-home-care services and multidisciplinary care in PSP and APD.

Patients' data	PSP N = 129	APD N = 65	p
In-home-care services			
Day-time assistance, days/year	291 (123)	6.1 (1.5)	**<0.001**
Night-time assistance, days/year	315 (117)	7.5 (0.5)	**<0.001**
At least one in-home care service, n (%)	28 (21.7)	18 (27.6)	0.22
Number of patients with at least one service over the previous year:			
Nurse, n (%)	12 (9.3)	5 (7.6)	0.46
Day-time assistance, n (%)	21 (16.3)	14 (21.5)	0.38
Night-time assistance, n (%)	8 (6.2)	4 (6.1)	0.62
Meal delivery, n (%)	6 (4.7)	1 (1.5)	0.25
Home structural changes, n (%)	42 (32.6)	9 (13.8)	**0.03**
Social assistant, n (%)	4 (3.1)	4 (6.1)	0.22
Transport assistance, n (%)	10 (7.8)	6 (9.2)	0.45
Multidisciplinary care visits,% of patients with at least one visit over the previous year			
Neurologist	98.4	97.3	0.46
GP	99	100	1
Geriatrician	19.4	13.2	0.22
Physical and Rehabilitation Medicine	51.2	38.8	0.06
Physiotherapist	62.7	50.7	**0.05**
SLT	33.3	15.3	**0.006**
Occupation Therapist	4.7	0	0.08
Psychologist	26.1	7.6	0.18
Psychiatrist	9.3	10.8	0.45
BNT	5.4	4.1	0.43
Social assistance	6.2	6.8	0.64
Surgeon	12.4	12.2	0.57
Dietician	13.2	8.1	0.29
Pulmonologist	8.5	5.4	0.20
Gastroenterologist	3.1	13.8	**0.01**
Ophthalmologist	49.6	39.2	0.06
Urologist	27.1	23.3	0.33
Spiritual care	3.1	2.7	0.69

Abbreviations: APD: Advanced Parkinson's disease; BNT: botulinum toxin treatment; GP: general practitioner; PSP: Progressive Supranuclear Palsy; PT: physiotherapist; SLT: speech and language therapist. Data are expressed in mean (standard deviation) unless otherwise specified.

hours of assistance (p = 0.005 and p = 0.080, respectively).

Looking at regional differences, no demographic and clinical differences were found comparing patients belonging to the Northern, Center and Southern part of Italy (data not shown).

3.2. Progressive supranuclear palsy: in-home care services and multidisciplinary interventions

In-home care use and multidisciplinary care for PSP patients are summarized in Table 2 and Table S1, S2 and S3. Overall, 21.7 % of PSP patients had at least one in-home care intervention, with 32.6 % of the patients having required home structural changes. Only 9.3 % of the patients benefited of visits from a home nurse and 3.1 % met a social assistant. Those who benefited from daytime or night-time home assistance (16.3 % and 6.2 %, respectively) had it for most of the year (291 and 315 days/year). Regarding multidisciplinary interventions, almost all patients had met at least one time over the past year the GP as well as the neurologist. More than 60 % of PSP had a visit with physiotherapist and about half met the physical and rehabilitation physician and the ophthalmologist. Other specialists often consulted were geriatricians, SLT, psychologists and urologists (Table 2). The highest number of visits per year was performed with physiotherapist and SLT (23 and 7, respectively, Table S1). Conversely, OT, psychiatrist, BNT, social and spiritual assistance, pulmonologist, gastroenterologist were consulted in a limited number of patients (Table 2). Most of the multidisciplinary care was performed within the NHS in the majority of cases (range 70–100 %) with the following exceptions: physiotherapists (56 %), occupational therapist (66 %), pulmonologist (54 %), gastroenterologist (60 %), ophthalmologist (53 %), urologist (56 %), home care nurse visits (58 %) home care day time assistance (9 %). Home care night-time assistance was never provided by the NHS (0 %) (Table S2).

Looking at regional differences for in-home care services (Northern versus Central versus Southern Italy), no major discrepancies were highlighted except for a higher number of structural changes for PSP patients belonging to Northern Italy if compared to other regions (p = 0.008). As for multidisciplinary care visits patients from the Southern Italy were more likely to visit an ophthalmologist but less likely to meet a SLT (p = 0.03 and p = 0.005, respectively) (Table S1).

3.3. Progressive supranuclear palsy patients: risk factors for in-home care and multidisciplinary interventions

At the univariate logistic analysis the following clinical and social factors resulted to be significant: a) female sex (p = 0.016), older age (p = 0.017), clinical milestones (gait [p = 0.002], cognitive impairment [0.006], dysarthria [p = 0.022]), higher PSPRS total score (p < 0.001), and lower S&E (p = 0.001), for the presence of a day-time assistance; b) one clinical milestone (dysarthria [p = 0.022]), higher PSPRS total score (p = 0.033) for the need of at least one in-home care service; c) higher PSPRS total score (p = 0.003), a lower S&E (p = 0.04), one clinical milestone (dysphagia [p = 0.031]) and the PSP-RS variant (p = 0.008) for the realization of home structural changes. No variable was associated to performing more than 4 multidisciplinary visits/year. No one of

the variables that resulted to be significant at the univariate analysis, kept its significance at the multivariate analysis.

3.4. Progressive supranuclear palsy versus advanced Parkinson's disease

Regarding in-home care services, PSP patients needed more days/year of day-time and night-time assistance ($p < 0.001$) as well as more frequent home structural changes ($p = 0.03$) if compared to APD (Table 2, Fig. 1).

A few differences were also found for multidisciplinary care. Indeed, a higher proportion of PSP patients have met at least once a SLT ($p = 0.006$). There was a trend towards significance for a greater proportion of PSP with at least one visit with physiotherapist for PSP ($p = 0.05$), while APD consulted more frequently at least once a gastroenterologist ($p = 0.01$) (Table 2, Fig. 1). However, the mean number of visits with physiotherapist was higher for APD if compared to PSP patients ($p < 0.001$), while the mean number of different physicians met over the previous year was higher for PSP patients ($p = 0.01$) (Table S1).

3.5. Global risks factors for in-home care needs and multidisciplinary interventions

When considering both PSP and APD patients as a unique population, we found several demographic and clinical variables as determinants for in-home care or multidisciplinary interventions needs. At the univariate logistic analysis the following clinical and social factors resulted to be significant: a) female sex ($p = 0.035$), older age ($p = 0.003$), living without a family caregiver ($p < 0.001$), and lower S&E score ($p < 0.001$), for the presence of a day-time home care assistance; b) longer disease duration ($p = 0.015$), lower S&E score ($p = 0.018$), and living without a family caregiver ($p = 0.04$) for the need of at least one in-home care service; c) PSP diagnosis ($p = 0.002$) and belonging to a Northern Italy center of care ($p < 0.001$) for the realization of home structural changes; d) PSP diagnosis ($p = 0.01$) for having done at least 4 multidisciplinary visit over the previous year.

At multivariate logistic regression analysis: a) lower S&E score (OR: 0.95; 95 % CI: 0.93–0.97; $p < 0.001$) and living without a family caregiver (OR: 4.7; 95 % CI: 1.88–11.60; $p = 0.001$) kept their significance for the presence of a day-time home care assistance; b) lower S&E score (OR: 0.98; 95 % CI: 0.96–0.99; $p < 0.04$) and living without a family caregiver (OR: 2.8; 95 % CI: 1.28–6.50; $p = 0.01$) kept their significance

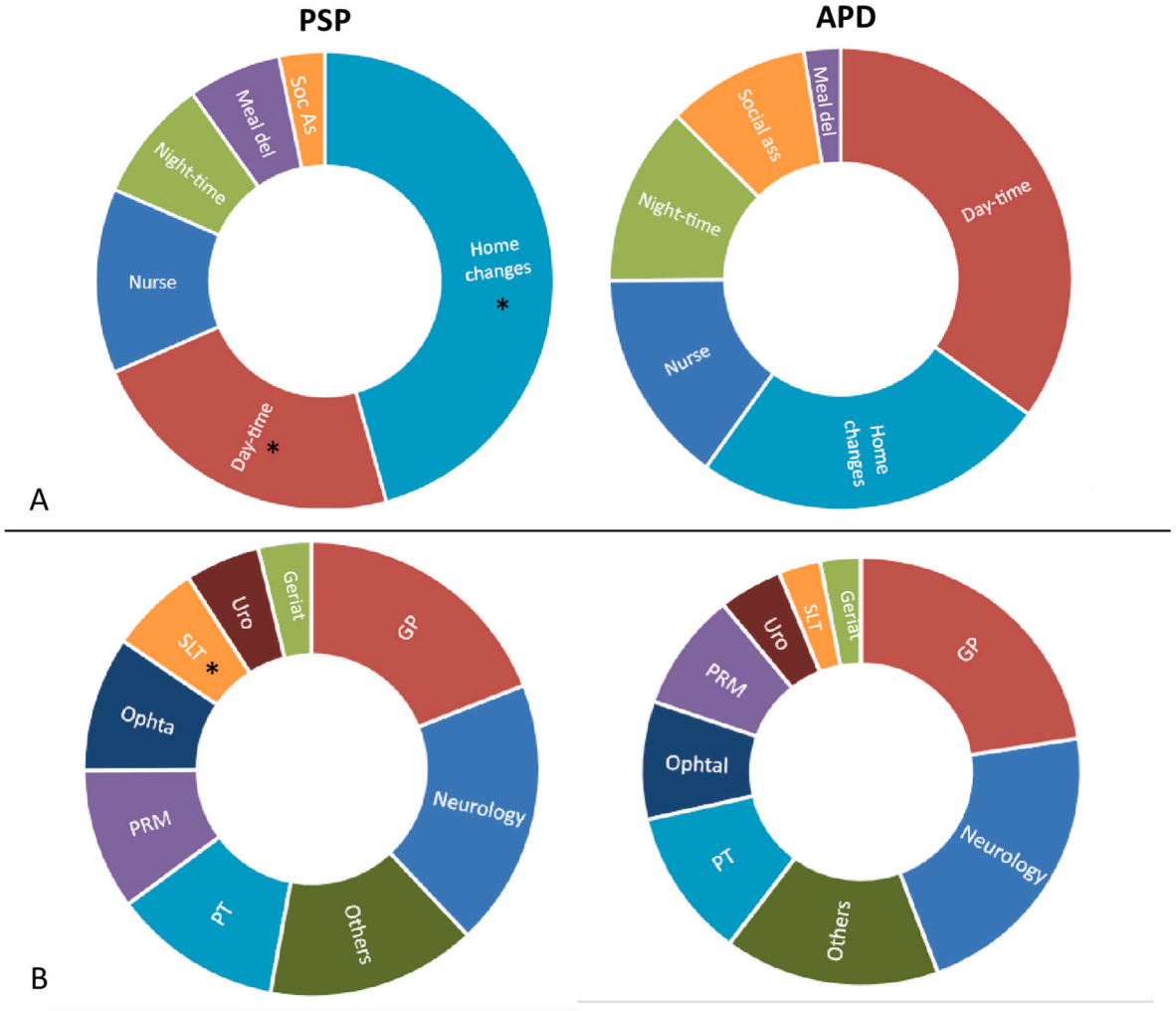

Fig. 1. Distribution of in-home care services (A) and multidisciplinary visits (B) (percentage of patients with at least one visit over the previous year) in PSP and APD. (*) indicate the statistically significant differences between groups ($p < 0.05$). Multidisciplinary care interventions appear with individual slice in the graphic if the % was at least 3 %.

Abbreviations: APD: advanced Parkinson's disease; geriatr: geriatrician; GP: general practitioner; Meal del: meal delivery; ophtal: ophthalmologist; PRM: physical and rehabilitation physician; PSP: progressive supranuclear palsy; PT: physiotherapist; SLT: speech and language therapist; Soc As: Social assistance; Uro: urologist.

for the need of at least one home care service; c) PSP diagnosis (OR: 3.07; 95 % CI: 1.39–6.80; $p = 0.006$) and belonging to a Northern Italy center of care (OR: 5.2; 95 % CI: 2.02–12.69; $p < 0.001$) kept their significance for the realization of home structural changes; d) PSP diagnosis (OR = 2.25; 95 % CI: 1.18–4.30; $p = 0.01$) for having done at least 4 multidisciplinary visit over the last year.

4. Discussion

Herein, we have performed a real-life multicentre cohort study on a large population of PSP patients to evaluate the pattern of use of in-home care services and multidisciplinary interventions across Italy. About one fifth of the patients had at least one in-home care intervention. GPs, neurologists, physiotherapists, ophthalmologists, physical and rehabilitation physicians, geriatricians, SLTs, psychologists and urologists were the most consulted healthcare professionals, in most cases within the NHS. We did not disclose clinical or social variables related to higher multidisciplinary or home care needs that survived at the multivariate regression analysis for PSP patients. Conversely, when considering APD and PSP as a unique group, both lower S&E and living without a familial caregiver were related to higher home care needs. Furthermore, within such a model, a PSP diagnosis was related to home structural changes and more than 4 multidisciplinary care visits, suggesting a potential requirement for tailored in-home adaptations and a multidisciplinary approach for PSP patients.

Indeed, while the literature on multidisciplinary care for PD patients is quite vast, for atypical parkinsonism there is a dearth of knowledge in the field [2,13,19]. To fill this gap, in USA and Canada, the CurePSP Center of Care network was established in 2017, aiming to improve early diagnosis but also optimize standard of care for patients with atypical parkinsonism [20]. Based on the few evidence available, clinicians suggest PSP patients to benefit of physiotherapy and weighted walkers to decrease the risk of falls and to perform SLT to cope with vocal changes and chocking [21–23]. A few patients are also referred to occupational therapy for home adaptations as well as to palliative care consultants for nursing home placement, end life decision including caregivers. Yet, a large, randomized trial including patients with PD and with atypical parkinsonism recently showed the benefit of an outpatient integrated palliative care approach performed by neurologists, social workers, chaplains, and nurses on quality of life and caregiver burden [3]. However, little is known about the feasibility in real-life clinical settings of multidisciplinary and palliative care in PSP [24].

Our cohort study highlights the existing gaps for an effective multidisciplinary care in real-life clinical settings in Italy. As a matter of fact, about 40 % of PSP patients has not met a physiotherapist over the previous year, only one third met a SLT and about 5 % an occupational therapist. Likewise, home care services are not extensively implemented (range 2–16 %), except for home structural changes (32.6 %), especially for patients from the Northern part of the country. We speculate this territorial disparity may be linked to higher individual income in Northern Italy compared to Southern Italy, as home structural changes due to health reasons are rarely supported by the NHS. When looking at predictors of in-home care or multidisciplinary care needs, we found a significant association within the univariate models for greater global disease severity (PSPRS total score), reduced functional autonomy (S&E) and the presence of clinically meaningful milestones of disease (gait and cognitive impairments, dysarthria and dysphagia) as well as the PSP-RS phenotype. However, none of those variables was confirmed within the multivariate analysis. Nevertheless, as a matter of fact in real-life clinical settings all those factors help the clinician in best targeting the most disabled patients and need to be kept into account in a potential care planning for PSP. Among those factors, the PSP-RS phenotype is well known to represent a marker of faster deterioration of disease compared to other phenotypes [25].

As further objective of our study, we performed a case-control study to compare the complex care use of a rare neurodegenerative condition, i.e. PSP, with a common neurodegenerative condition, i.e. PD, though targeting the advanced-late phase of the latter. The primary aim of this case-control sub-analysis was to verify if PSP patients had a reduced access to in-home services or multidisciplinary care. To verify this hypothesis, we decided to choose as controls, PD patients with an already defined axial impairment (H&Y ≥ 3). Of note the selected group of APD patients had a lower S&E and a higher neuropsychiatric burden, mainly related to hallucinations, mood disorders and sleep problems. Comparing the use of in-home care and multidisciplinary visits, we failed to find major differences, indicating that PSP needs are like APD ones. Indeed, PSP manifest such needs in about two years of disease duration, while for PD such time is 6-folded. As a matter of fact, the access to the health services for PSP needs to be planned promptly and since the earliest phases of the disease.

When we considered both groups as a unique population, we found a few clinical and social elements significantly related to more complex care needs, such as lower S&E, living without a familial caregiver and having a PSP diagnosis. The S&E is universally used to define the autonomy of patients. A score lower than 50 % has already been suggested to be indicated to identify late-stage PD patients and we confirm its utility in identifying patients with complex care needs also in PSP [26].

An interesting finding of our study is the extremely low proportion of both PSP and APD patients who have discussed their living will. This highlights a gap between the real-life clinical practices and the literature recommendations. In 2020 a systematic review looked for experiences in advance care planning for PD and atypical parkinsonism and only four studies have focused on this issue in atypical parkinsonism [4]. Overall, a lack of knowledge of disease progression and/or palliative care approaches was reported amongst non-specialist health-care professionals, along with the fact that patients felt it was often left to them to initiate such discussion. Interestingly, one study including 29 people with different neurological conditions (dementia, PD, Huntington's disease, PSP, motor neurone disease, multiple sclerosis), found out that PSP patients were better informed about the prognosis of their condition and about possible decisions, e.g. on percutaneous endoscopic gastrostomy feeding, compared to people with PD [27]. The discussion of advance care planning is likely not systematically discussed by physicians with patients with neurodegenerative diseases, even if cultural disparities among different countries need to be considered [28]. However, we acknowledge in depth evaluation of advance care planning was out of the scope of this sub-study of the PSP NET.

The present study is the result of a multicenter survey conducted at a national level and may represent a window to describe the real-world situation of care ongoing in high-income countries with national-based health system as Italy. It is noteworthy that none of the movement disorder centers included in the study had access to an advanced practice nurse, a role that plays a key part in the management of neurodegenerative diseases [29–31]. While regrettable, this situation remains the reality for many European countries. At the same time, our study can be the starting point to identify pros and cons of the health services offered to patients with atypical parkinsonism to move forward to build an integrated, patient-centered model of care for such patients [32].

Our study has limitations that should be considered for the interpretation of the results. Although we have targeted a large PSP population, we missed to evaluate also related caregivers burden and the subjective perceived needs of patients as well as patient/family income. Additionally, we did not evaluate cognitive impairment which could have had a specific impact on complex care needs. However, the S&E is usually correlated to the presence of cognitive impairment as well as the H&Y and they have been both considered in our analysis.

In conclusion, we offer a real-life screenshot of the current in-home care use and multidisciplinary interventions in a large cohort of PSP patients in Italy. We highlight the existing gaps to reach an efficient multidisciplinary care for this rare disease as about 40 % of PSP patients has not met a physiotherapist over the previous year, only one third met a SLT and about 5 % an occupational therapist. A low level of functional

autonomy, a PSP-RS phenotype and higher PSPRS score may be considered as red flags for higher complex care needs. When compared to APD patients, PSP seems to need a higher frequency of home structural changes. Our findings claim for the need to establish a shared integrated care planning at a national level to be proposed promptly to patients affected by atypical parkinsonism.

Funding sources and conflict of interest

This research is supported by the Fondazione LIMPE per il Parkinson Onlus who supports the PSP-NET project.

The authors have no conflict of interest for this study.

Financial disclosures for the previous 12 months

Margherita Fabbri received Honoraria to speak from AbbVie, ORKYN, and BIAL, consultancies for BIAL, LVL Médical and Convatec; Grants from: MSA Coalition and HORIZON 2022.

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The other authors have no financial disclosures for the past 12 months.

Ethical compliance statement

The project was approved by the Ethical committee of the coordinating center (number 178 December the 4th, 2020) and, thereafter, by the ones of all participating centers.

CRediT authorship contribution statement

Margherita Fabbri: Writing – review & editing, Writing – original draft, Conceptualization. **Claudia Ledda:** Writing – review & editing, Investigation. **Tommaso Schirinzi:** Writing – review & editing, Investigation. **Carlo Alberto Artusi:** Writing – review & editing, Investigation. **Anna Rosa Avallone:** Writing – review & editing, Investigation. **Henri Zenuni:** Writing – review & editing, Investigation. **Rosa De Micco:** Writing – review & editing, Investigation. **Simone Aloisio:** Writing – review & editing, Investigation. **Ilaria Cani:** Writing – review & editing, Investigation. **Maria Chiara Malaguti:** Writing – review & editing, Investigation. **Francesca Di Biasio:** Writing – review & editing, Investigation. **Giovanna Calandra-Buonaura:** Writing – review & editing, Investigation. **Alessandro Stefani:** Writing – review & editing, Investigation. **Leonardo Lopiano:** Writing – review & editing, Investigation. **Paolo Barone:** Writing – review & editing, Investigation. **Marina Picillo:** Writing – review & editing, Writing – original draft, Conceptualization.

Declaration of competing interest

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None of the authors report any financial interests or professional relationship related to the subject matter but not directly to this manuscript for the last 3 years.

There are no patents or copyrights licensed to the authors that are relevant to the work submitted for publication.

There are no additional relationships or activities to declare that may be perceived to have influenced the submitted work.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.parkreldis.2024.107047.

Appendix 1

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References

[1] R. Rajan, L. Brennan, B.R. Bloem, et al., Integrated care in Parkinson's disease: a systematic review and Meta-analysis, Mov. Disord. 35 (9) (2020) 1509–1531.
[2] D.L.M. Radder, J. Nonnekes, M. van Nimwegen, et al., Recommendations for the organization of multidisciplinary clinical care teams in Parkinson's disease, J. Parkinsons Dis. 10 (3) (2020) 1087–1098.
[3] B.M. Kluger, J. Miyasaki, M. Katz, et al., Comparison of integrated outpatient palliative care with standard care in patients with Parkinson disease and related disorders: a randomized clinical trial, JAMA Neurol. 77 (5) (2020) 551–560.
[4] D. Nimmons, L. Hatter, N. Davies, E.L. Sampson, K. Walters, A. Schrag, Experiences of advance care planning in Parkinson's disease and atypical parkinsonian disorders: a mixed methods systematic review, Eur. J. Neurol. 27 (10) (2020) 1971–1987.
[5] M. Fabbri, L.A. Kauppila, J.J. Ferreira, O. Rascol, Challenges and perspectives in the management of late-stage Parkinson's disease, J. Parkinsons Dis. 10 (s1) (2020) S75–s83.
[6] D.G. Coughlin, I. Litvan, Progressive supranuclear palsy: advances in diagnosis and management, Park. Relat. Disord. 73 (2020) 105–116.
[7] N. O'Shea, S. Lyons, S. Higgins, S. O'Dowd, Neurological update: the palliative care landscape for atypical parkinsonian syndromes, J. Neurol. 270 (4) (2023) 2333–2341.
[8] Late (Complicated) Parkinson's Disease. European Handbook of Neurological Management, Volume vol. 1.
[9] G.U. Höglinger, G. Respondek, M. Stamelou, et al., Clinical diagnosis of progressive supranuclear palsy: the movement disorder society criteria, Mov. Disord. 32 (6) (2017) 853–864.
[10] R.B. Postuma, D. Berg, M. Stern, et al., MDS clinical diagnostic criteria for Parkinson's disease, Mov. Disord. 30 (12) (2015) 1591–1601.
[11] M.M. Hoehn, M.D. Yahr, Parkinsonism: onset, progression and mortality, Neurology 17 (5) (1967) 427–442.
[12] S.T. Jost, M.A. Kaldenbach, A. Antonini, et al., Levodopa dose equivalency in Parkinson's disease: updated systematic review and proposals, Mov. Disord. 38 (7) (2023) 1236–1252.
[13] M.A. Qamar, G. Harington, S. Trump, J. Johnson, F. Roberts, E. Frost, Multidisciplinary care in Parkinson's disease, Int. Rev. Neurobiol. 132 (2017) 511–523.
[14] J.L. Cummings, M. Mega, K. Gray, S. Rosenberg-Thompson, D.A. Carusi, J. Gornbein, The Neuropsychiatric Inventory: comprehensive assessment of psychopathology in dementia, Neurology 44 (12) (1994) 2308–2314.
[15] A.C. Schwab Rse, in: E. And, S. Livingstone (Eds.), Third Symposium on Parkinson's Disease, Sage, Edinburgh, UK, 1969, pp. 152–157.
[16] L.I. Golbe, P.A. Ohman-Strickland, A clinical rating scale for progressive supranuclear palsy, Brain : J. Neurol. 130 (Pt 6) (2007) 1552–1565.
[17] A. Antonini, G. Abbruzzese, L. Ferini-Strambi, et al., Validation of the Italian version of the movement disorder society–unified Parkinson's disease rating scale, Neurol. Sci. : official journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology 34 (5) (2013) 683–687.
[18] M.J. Grimm, G. Respondek, M. Stamelou, T. Arzberger, L. Ferguson, E. Gelpi, et al., How to apply the movement disorder society criteria for diagnosis of progressive supranuclear palsy, Mov. Disord. 34 (8) (2019 Aug) 1228–1232.
[19] D.L.M. Radder, N.M. de Vries, N.P. Riksen, et al., Multidisciplinary care for people with Parkinson's disease: the new kids on the block, Expert Rev. Neurother. 19 (2) (2019) 145–157.
[20] B. Bluett, A.Y. Pantelyat, I. Litvan, F. Ali, D. Apetauerova, D. Bega, et al., Best practices in the clinical management of progressive supranuclear palsy and corticobasal syndrome: a consensus statement of the CurePSP centers of care, Front. Neurol. 12 (2021) 694872.
[21] I. Clerici, D. Ferrazzoli, R. Maestri, et al., Rehabilitation in progressive supranuclear palsy: effectiveness of two multidisciplinary treatments, PLoS One 12 (2) (2017) e0170927.
[22] C. Zampieri, R.P. Di Fabio, Balance and eye movement training to improve gait in people with progressive supranuclear palsy: quasi-randomized clinical trial, Phys. Ther. 88 (12) (2008) 1460–1473.
[23] P. Sale, D. Castiglioni, M.F. De Pandis, et al., The Lee Silverman Voice Treatment (LSVT®) speech therapy in progressive supranuclear palsy, Eur. J. Phys. Rehabil. Med. 51 (5) (2015) 569–574.
[24] E. Tilley, J. McLoughlin, S.A. Koblar, et al., Effectiveness of allied health therapy in the symptomatic management of progressive supranuclear palsy: a systematic review, JBI database of systematic reviews and implementation reports 14 (6) (2016) 148–195.
[25] A. Shoeibi, I. Litvan, E. Tolosa, T.D. Ser, E. Lee, Progression of two Progressive Supranuclear Palsy phenotypes with comparable initial disability, Park. Relat. Disord. 66 (2019) 87–93.
[26] M. Coelho, J.J. Ferreira, Late-stage Parkinson disease, Nat. Rev. Neurol. 8 (8) (2012) 435–442.
[27] G. Clarke, E. Fistein, A. Holland, J. Tobin, S. Barclay, S. Barclay, Planning for an uncertain future in progressive neurological disease: a qualitative study of patient and family decision-making with a focus on eating and drinking, BMC Neurol. 18 (1) (2018) 115.
[28] G. Saranza, E.Q. Villanueva 3rd, A.E. Lang, Preferences for communication about end-of-life care in atypical parkinsonism, Mov. Disord. 36 (9) (2021) 2116–2125.
[29] H.D. Lum, S.R. Jordan, A. Brungardt, R. Ayele, M. Katz, J.M. Miyasaki, A. Hall, J. Jones, B. Kluger, Framing advance care planning in Parkinson disease: patient and care partner perspectives, Neurology 92 (22) (2019) e2571–e2579.
[30] D. Nimmons, L. Hatter, N. Davies, E.L. Sampson, K. Walters, A. Schrag, Experiences of advance care planning in Parkinson's disease and atypical parkinsonian disorders: a mixed methods systematic review, Eur. J. Neurol. 27 (10) (2020) 1971–1987.
[31] S. Thomas, E. Edwards, C. Kobylecki, Parkinson's nurses are crucial for the management of Parkinson's disease: 2007-2024, J. Parkinsons Dis. (2024) 1–9, https://doi.org/10.3233/JPD-230224. Published online April 5.
[32] B.R. Bloem, E.J. Henderson, E.R. Dorsey, et al., Integrated and patient-centred management of Parkinson's disease: a network model for reshaping chronic neurological care, Lancet Neurol. 19 (7) (2020) 623–634.