


# BMJ Open Impact of severe polyhandicap cared for at home on French informal caregivers' burden: a cross-sectional study

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**To cite:** Rousseau M-C, Baumstarck K, Valkov M, *et al.* Impact of severe polyhandicap cared for at home on French informal caregivers' burden: a cross-sectional study. *BMJ Open* 2020;**10**:e032257. doi:10.1136/bmjopen-2019-032257

► Prepublication history and additional material for this paper are available online. To view these files, please visit the journal online (<http://dx.doi.org/10.1136/bmjopen-2019-032257>).

Received 11 June 2019  
Revised 22 November 2019  
Accepted 02 December 2019



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## ABSTRACT

**Objectives** Polyhandicap (PLH), defined by a combination of profound intellectual impairment and serious motor deficits, is a severe condition with complex disabilities. In France, care of the large majority of PLH individuals is managed in specialised rehabilitation centres or residential facilities, but some of PLH individuals are cared for at home. The aims of this study were to assess the self-perceived burden among informal caregivers of PLH individuals and to identify potential determinants of this burden.

**Design** Cross-sectional study (Neuropaediatric Department, Trousseau Hospital, Paris, France).

**Settings** PLH children were recruited from a specialised paediatric/neurological department.

**Participants** The selection criteria of caregivers were age above 18 years and being the PLH individual's next of kin.

**Outcomes measures** From March 2015 to December 2016, data were collected from the caregivers, including sociodemographical data, health status, psycho-behavioural data (quality of life, mood disorders and coping) and self-perceived burden. In addition, the health status of the PLH individual was collected. Relationships between the burden scores and potential determinants were tested (correlations coefficients, Mann-Whitney tests, generalised estimating equations models).

**Results** Eighty-four children were eligible; 77 families returned their questionnaire. The informal caregivers of PLH children experienced a high level of perceived burden (scores ranged from 55±20 to 81±12). Eighty per cent of them had more than 5 hours of daily caregiving and 51% of them had to get up more than twice during the night. The main factors associated with caregiver burden were age, financial issues, health status, daily care and coping strategies. The patients' health status was not associated with caregiver burden.

**Conclusions** Some of the caregiver burden determinants might be modifiable. These findings should help healthcare workers and health-decision makers implement specific and appropriate interventions.

**Trial registration number** NCT02400528.

## BACKGROUND

Polyhandicap (PLH) is a severe health condition with complex disabilities. This chronic disorder occurs in an immature brain,

## Strengths and limitations of this study

- This study explored, for the first time, the burden on the informal caregivers of polyhandicap (PLH) individuals cared for at home.
- This study was performed on a large sample allowing a high representativeness.
- These findings, based on a cross-sectional design, should be confirmed by longitudinal approaches.
- Based on a small sample size; our results may benefit from confirmation in a larger sample.
- Qualitative studies should be performed to better understand the determinants of PLH parents' burden.

leading to profound intellectual impairment and serious motor deficit. The result is an extreme restriction of autonomy and communication. The definition of PLH was adopted by the French scientific community and the French law (French Law no. 89-798, 27 October 1989, health policy of disability care).<sup>1,2</sup> PLH is similar to profound intellectual and multiple disabilities, but it does not systematically refer to a disorder affecting an immature brain.<sup>3</sup> In France, the prevalence of PLH is estimated in the paediatric population to be between 0.7 and 1.28 per thousand, and 880 new cases are diagnosed per year.<sup>4</sup> Due to the various serious disabilities and multiple comorbidities,<sup>5</sup> PLH patients need permanent human and technical assistance throughout their life.

The French health system allows these individuals to benefit from two main care-management modalities: specialised re-education centres, providing a high level of medical care for patients with the most severe health issues, and residential facilities, offering more educational care for less severely affected individuals.<sup>6</sup> However, the care of some individuals, mainly children or young adults, is managed at home. Due to the improvement in life expectancy, the

progressive move from traditional institutional care to deinstitutionalisation care, and a (slow) reduction in stigmatisation, family caregivers assume new roles that were previously carried out by institutional caregivers. Families of PLH children, from birth or in the first years of life of the individual, are repeatedly and chronically confronted with major nursing care and stressful situations. Despite being supported by human and technical assistance, the burden on families is likely to be considerable. Some studies have explored burden on informal caregivers who have children with multiple disabilities<sup>7–9</sup> and in informal caregivers who have children with cerebral palsy.<sup>10–14</sup> The caregivers reported deteriorated health status, financial difficulties, restriction in their social participation and physical overload. Limited data are available for caregivers of patients with PLH.<sup>15 16</sup> However, an assessment of parent burden and knowledge of which factors are determinants of this burden would strongly assist clinicians and health decision-making authorities in offering appropriate interventions such as human and technical assistance, respite and supportive care, and psychological support.

The aims of this study were as follows: (i) to describe the health profile of PLH individuals whose care is managed at home, (ii) to assess the burden on their informal caregivers and (iii) to identify potential determinants of caregiver burden.

## METHODS

### Design and settings

This study was based on a cross-sectional design and was included in the French national PLH study. The general aim of the French PLH study was to identify the potential (socioeconomical, environmental and epidemiological) determinants of the health status of PLH individuals and understand the daily life of their informal caregivers. PLH individuals with care managed at home were recruited from a specialised paediatric/neurological department of a university hospital (Neuropaediatric Department, Trousseau Hospital, Assistance Publique Hôpitaux de Paris, France).

### Selection criteria

Eligible criteria of PLH individuals were severe PLH defined by the combination of motor deficiency (tetraparesis, hemiparesis, paraparesis, extrapyramidal syndrome, cerebellar syndrome and neuromuscular problems) and intellectual impairment ( $IQ < 40$ ), associated with everyday life dependence (functional independence measure (FIM)  $< 55$ ), restricted mobility (gross motor function classification system (GMFCS) levels III, IV and V), age at onset of cerebral lesion under 3 years old and care managed at home. The selection criteria of informal caregivers were as follows: aged above 18 years, the PLH individual's next of kin and voluntary consent to participate.

### General procedure

For each PLH individual included in the study, data were collected from medical records obtained by a dedicated

clinical research assistant and were checked by the patient's referent physician. Next of kin (individuals to contact for any medical, administrative and social issues) were systematically identified in the medical record. From 19 March 2015 to 31 December 2016, a booklet, including self-administered questionnaires, was sent by mail to each next of kin. To optimise participation, a prepaid return envelope addressed to the coordination team send with the booklet.

### Data collection

Two sources of data were used: medical records and caregiver data.

1. Patient characteristics were collected from the medical records:
  - Sociodemographics: age and gender.
  - Aetiology: unknown versus known, progressive versus non-progressive disease.
  - Global health severity: severe for patients who met all the following criteria: motor handicap (paraparesia or tetraparesia and/or extrapyramidal syndrome and/or severe general hypotonia),  $IQ < 25$ ,  $FIM \leq 20$ , and GMFCS IV and V; less severe for patients who did not meet these criteria.
  - Global health stability: unstable for patients who met at least one of the following criteria: recurrent pulmonary infections ( $\geq 5$ /year), drug-resistant epilepsy ( $\geq 4$  seizures/month); stable for patients who did not meet any of these criteria.
  - Medical devices: at least one device (including invasive mechanical ventilation, non-invasive mechanical ventilation, tracheotomy, nasogastric tube, gastrostomy, permanent urinary catheter, cerebrospinal fluid derivation and central venous catheter).
  - Usual means of care: home care, specialised re-education centre or residential facility.
2. Data collected from caregivers were recorded in a booklet, and included the following:
  - Sociodemographical and professional status: age, gender, nature of the relationship with the patient, marital status (not single/single), in a couple with the patient's other parent, number of children, another disabled individual living at home, educational level, occupational status (employed/unemployed) and self-perceived financial status.
  - Health: chronic diseases, hospitalisation episode during the last 2 years.
  - Psycho-behavioural characteristics: (i) quality of life (QoL): assessed using the WHO Quality of Life (WHOQoL-BREF, brief version of the WHO-QoL-100) questionnaire, a generic questionnaire used worldwide describing four domains: physical health, psychological health, social relationships and environment. All scores ranged between 0 and 100. Higher scores indicated a better QoL; (ii) anxiety-mood disorders, assessed using a Likert Scale ranging from 1 (absence) to 10 (very significant); (iii) coping, assessed using the Brief Coping Ori-

entation to Problems Experienced Scale, exploring four dimensions that include social support, problem solving, avoidance and positive thinking.<sup>17</sup> Scores ranged from 0 to 100. High scores reflected a high tendency to implement the corresponding coping strategies.

- Burden: (i) specific ad hoc questions: number of children living at home, hours of daily caregiving and frequency of getting up during the night; (ii) self-perceived burden using the Caregiver Reaction Assessment (CRA) Scale,<sup>18</sup> a 24-item questionnaire describing five subscales: self-esteem (seven items), family support (five items), financial impact (three items), planning impact (five items) and health impact (four items). Scores ranged from 1 to 5. For the self-esteem subscale, a higher score indicated that caregiving had a more positive impact; a score  $\geq 4$  was used to indicate a high positive impact. For the other four subscales, higher scores indicated that caregiving had a more negative impact; a score  $\geq 3$  was used to indicate a high negative impact. All the scores were linearised on a 0–100 scale.

Additional information was collected and detailed in the online supplementary file (additional table 1).

### Consent to participate

A written consent form was obtained for each participant.

### Statistical analysis

The CRA scores were calculated according to the tool's scoring rules. Comparison of CRA scores between different subgroups (parent variables: gender, nature of the relationship with the patient, marital status, other disabled individuals at home, educational level, occupational status, financial status, chronic disease(s), hospitalisations, hours of daily caregiving, getting up during the night; patient variables: gender, severity, stability, medical devices) was performed using Mann-Whitney tests. Associations between CRA scores and continuous variables (age of parents, number of children, QoL scores, anxiety–mood score, coping scores, number of children at home, age of children) were analysed using Spearman's correlations. Multivariate analyses using generalised estimating equations models were performed to identify variables linked to CRA scores. In the models, each CRA score was considered to be a separate dependent variable. The independent variables relevant to the models were selected from the univariate analysis, based on a threshold p value of  $\leq 0.05$ . The final models produced standardised beta coefficients, which represented the change in the SD of the dependent variable (CRA) resulting from a change of one SD in the various independent variables. Statistical analyses were performed using SPSS N°22 software (IBM SPSS PASW Statistics). All tests were two-sided. The threshold for statistical significance was set at  $p < 0.05$ .

### Patient and public involvement

Some parents of PLH patients were involved during the validation of the final data collection form to provide their opinions on the choice of questionnaires, understanding/formulation of the items, length of the form and so on. At the end of the study, a research clinical assistant contacted a sample of 50 non-respondents by phone to collect the reasons for non-participation (time consuming, language difficulties, avoidance and so on).

### RESULTS

A total of 84 children were eligible. Seventy-seven of the families returned their questionnaires (response rate: 92%).

#### Characteristics of the children

The youngest child was 3 years old and the oldest was 27 years. The gender ratio was 1. The PLH aetiology was unknown for 13% and progressive for 18%. Almost 70% of the children presented with severe PLH and 42% of them presented with unstable disease. The developmental level according to the Brunet-Lezine Scale was low: 3–6 months for all domains. Approximately 30% of PLH children presented with visual impairment and 27% presented with behavioural disorders. The most frequent comorbidities were epilepsy (52%), scoliosis (41%), gastro-oesophageal reflux (40%), drooling (32%), faecal impaction (31%) and aspiration syndrome (23%). The most prescribed medications were anticonvulsants, laxatives and osteoporotic prevention. Almost 30% had at least one medical device. Eighty-two per cent of patients spent more than 3 hours daily positioned in sitting devices, 20% spent more than 3 hours daily in a verticalisation device and 4% spent more than 3 hours daily on an air mattress. Twenty-three per cent of the children had been admitted previously to the intensive care unit or the emergency department, and 4% of them were in a palliative condition. All characteristics of the children are described in tables 1 and 2.

#### Characteristics of the informal caregivers

The mean (SD) age of the participants was 41 (10) years. Most of the participants were the parents, usually the mothers. Ten per cent of the participants reported living with a second disabled individual. Half of them were employed and 34% of them self-reported financial difficulties. Almost 30% of the respondents reported at least one chronic disease and 23% reported a hospitalisation episode during the last 2 years. For support and financial assistance, 83% of the informal caregivers received a disability allowance, 10% received a disabled facility grant, 50% received a technical assistance allowance and almost 20% received human assistance at home. Twenty-two per cent of caregivers received psychological support, 43% had a social worker and 26% were involved in a disability association. Thirty-seven per cent of caregivers reported having a regular leisure activity and 44% took vacations.

**Table 1** Main characteristics of the children

	N=77
	N (%)
<b>Gender</b>	
Boys	38 (49.4)
Girls	39 (50.6)
<b>Age (years)</b>	
M±SD	7.8±5.7
Med (IQR)	5 (4–11)
<b>Polyhandicap aetiology*</b>	
Unknown	10 (13.2)
Known	66 (86.8)
Progressive disease	14 (18.4)
Non-progressive disease	62 (81.6)
<b>Severity†</b>	
Less severe	24 (31.2)
Severe	53 (68.8)
<b>Stability‡</b>	
Stable	45 (58.4)
Unstable	32 (41.6)
<b>Medical devices</b>	
None	54 (70.1)
At least one§	23 (29.9)
<b>Usual care modality</b>	
Home care	68 (88.3)
Specialised re-education centre	8 (10.4)
Residential facility	1 (1.3)

\*One missing data.

†Severe case: association of motor handicap, IQ<25, FIM≤20 and GMFCS IV/V.

‡Unstable case: recurrent pulmonary infections and/or drug-resistant epilepsy.

§At least one of the following list: gastrostomy (21), non-invasive mechanical ventilation (5), cerebrospinal fluid derivation (2), tracheotomy (1), permanent urinary probe (0), nasogastric tube (0), invasive mechanical ventilation (0) and central venous catheter (0). FIM, functional independency measure; GMFCS, gross motor function classification system.

Almost 40% of families reported being satisfied with caregiver services and 37% of parents reported being somewhat satisfied with the medical information given. All the details are provided in [table 3](#) and in the online supplementary additional table 1.

### Burden on the informal caregivers

For 80% of informal caregivers, the hours of daily caregiving were more than 5 hours per day; 51% of the informal caregivers had to get up more than twice during the night to care for the PLH individual. All CRA scores are described in [table 3](#).

**Table 2** Detailed characteristics of the children

	N=77
	N (%)
	Med (IQR)
<b>1. Health status</b>	
<b>Neurodevelopment status (months)*</b>	
Language	5 (2–9)
Posture-motor ability	3 (2–10)
Coordination	6 (2–6)
Sociability	6 (2–9)
<b>Neurological handicaps</b>	
Severe dystonia	5 (7.5)
Global hypotonia	38 (52.1)
Extrapyramidal syndrome	13 (19.1)
Ataxia	5 (7.5)
<b>Sensorial disorders</b>	
Visual impairment	22 (28.9)
Hearing impairment	7 (9.2)
<b>Behavioural disorders†</b>	20 (27.0)
<b>Sleep disorders</b>	
Night wake up	20 (26.7)
Difficulties falling asleep	12 (16.2)
Short sleep	3 (4.2)
Inversion	4 (5.4)
<b>Epilepsia</b>	
Presence of epilepsy	40 (52.6)
At least one crisis per day	27 (67.5)
Previous status epilepticus	19 (24.7)
<b>Orthopaedic</b>	
Scoliosis	31 (41.3)
Limb deformation	25 (34.2)
Hip luxation	11 (14.3)
Arthrodesis, other surgery	12 (15.8)
<b>Pulmonary</b>	
Aspiration syndrome	18 (23.4)
Recurrent pulmonary infections	8 (10.4)
<b>Digestive</b>	
Drooling	25 (32.5)
Faecal impaction	24 (31.2)
Gastro-oesophageal reflux	31 (40.3)
<b>Urinary</b>	
Recurrent urinary tract infections	1
Urinal retention	2
<b>Cutaneous</b>	
Bedsore	1
<b>2. Treatments and rehabilitation</b>	
<b>Medications</b>	

Continued



**Table 2** Continued

	<b>N=77</b>
	<b>N (%)</b>
	<b>Med (IQR)</b>
More than 3	35 (45.4)
Total number	3 (2–7)
Laxatives	42 (54.5)
Anticonvulsants	48 (62.3)
Psychotropics	7 (9.1)
Osteoporosis prevention	15 (19.5)
Antispastics	12 (15.6)
Antidystonics	8 (10.4)
Devices‡	
No device	54 (70.1)
One device	18 (23.4)
Two devices	4 (5.2)
Four devices	1
≥3 hours/day in a sitting device	63 (81.8)
Verticalisation device use	15 (19.5)
Air mattress	11 (14.3)
3. Health pathway	
Previous emergency and/or ICU admission	18 (23.4)
Perception of referent physician	
Medical care inadequacy	7 (9.1)
Educational care inadequacy	12 (15.6)
Request for an admission in institution§	21 (27.3)
Life project structuration	54 (70.1)
Palliative condition	3 (3.9)

Med (IQR): median (IQR).

\*Neurodevelopmental status levels range from 0 to 24 months.

†Behavioural disorders including intermittent screaming and/or agitation and/or stereotypies and/or intermittent crying and/or self-aggressivity and/or hetero-aggressivity.

‡Including gastrostomy, non-invasive mechanical ventilation, cerebrospinal fluid derivation, tracheotomy, permanent urinary probe, nasogastric tube, invasive mechanical ventilation and central venous catheter.

§Registered on a waiting list and/or official request made by the French Department for Persons with Disabilities.

ICU, intensive care unit.

### Factors associated with the informal caregivers' burden scores

#### Relationships between self-perceived burden of caregivers and their characteristics

For each CRA dimension, the factors associated with burden were as follows: (1) worst self-esteem dimension: use of avoidance as a coping strategy; (2) worst familial support dimension: having a second disabled individual living at home, a previous hospitalisation episode, reports of lower QoL scores, reports of higher anxiety–mood disorder levels, less use of social support as a coping

**Table 3** Characteristics of the informal caregivers

	<b>N=77</b>
	<b>N (%)</b>
	<b>M±SD</b>
	<b>Med (IQR)</b>
<b>1. Sociodemographics</b>	
Age (years)	41.4±10.2
	42 (34–47)
Gender	
Men	28 (36.8)
Women	48 (63.2)
Nature of relationship with the PLH individual	
Parents	69 (89.6)
Others*	7 (9.1)
Marital status	
Not single	65 (84.4)
Single	11 (14.3)
Couple with the other parent of the PLH individual	57 (74.0)
Number of children	3 (2–4)
Other person with disability living home	8 (10.4)
Educational level	
<12 years	35 (45.5)
≥12 years	42 (54.5)
Occupational status	
Not worker	38 (49.4)
Worker	38 (49.4)
Self-perceived financial status	
Not difficult	49 (63.6)
Difficult	26 (33.8)
<b>2. Health</b>	
Personal chronic disease(s)	22 (28.6)
Hospitalisation episode during the last 2 years	18 (23.4)
<b>3. Psycho-behavioral characteristics</b>	
Quality of life (WHOQoL scores) (0–100)†	
Physical	61.5±19.3
Psychological	65.8±15.7
Social	60.8±19.5
Environmental	58.7±13.9
Anxiety–mood score (1–10)‡	
Mean±SD	5.9±2.4
Coping strategies (Brief-COPE scores) (0–100)§	
Social support	39.5±19.7
Problem solvings	57.8±22.18
Avoidance	19.4±11.5

Continued



Table 3 Continued

	N=77
	N (%) M±SD Med (IQR)
Positive thinking	53.9±20.2
<b>4. Burden</b>	N (%)
Number of children living at home	2 (2–3)
<b>Daily time caregiving (hours)</b>	
≤4	16 (20.8)
(5–8)	26 (33.8)
≥9	30 (39.0)
<b>Get up during night</b>	
Never	10 (13.0)
1	23 (29.9)
≥2	39 (50.7)
<b>Self-perceived reaction (CRA scores) impact (0–100)</b>	
Self-esteem¶	81.1±11.5
Familial support**	53.1±17.7
Financial**	59.4±21.7
Scheduling/planification**	69.7±18.4
Health**	55.1±19.9

\*Others: 2 aunts, 2 grand-parents, 1 sibling, 1 cousin, 1 extra-familial relationship.

†Higher scores: higher QoL level.

‡Higher score: higher anxiety/mood disorder.

§Higher scores: higher use of the strategy.

¶100: best self-esteem.

\*\*100: worse familial support, worse financial impact, worse planification impact, worse health impact.

Brief-COPE, Brief Coping Orientation to Problems Experienced Scale; CRA, caregiver reaction assessment; PLH, polyhandicap; WHOQoL, WHO Quality of Life.

strategy, reports of longer hours of daily caregiving and getting up more than once during the night; (3) worst financial dimension: being older, having a second disabled individual at home, reports of financial difficulties, having an individual with a chronic disease, reports of lower QoL score, less use of positive thinking as a coping strategy; (4) worst planning dimension: previous personal hospitalisation episode, reports of lower QoL scores, reports of higher anxiety–mood disorder levels, use of avoidance as coping strategy, reports of longer hours of daily caregiving and reports of getting up more than once during the night; (5) worst health dimensions: being older, having a personal chronic disease, reports of lower QoL scores, reports of higher anxiety–mood disorder levels, use of avoidance as coping strategy and reports of getting up more than once during the night. The details are described in [table 4](#).

### Relationships between self-perceived burden on caregivers and the PLH individual's characteristics

Age, gender, severity and stability of the disease, and medical devices did not modulate the self-esteem, financial and planning dimensions. The older the PLH individual was, the worse the familial and the health scores were. Family support was worst when the PLH individual was male. Details are described in [table 5](#). Additional links are provided in online supplementary additional table 2.

### The most important predictors of burden

After adjustment, the factors associated with CRA scores were as follows: (1) worst familial support dimension: less use of social support as a coping strategy, getting up more than once during the night and older age of the PLH person; (2) worst financial dimension: having a second disabled individual at home and lower QoL score; (3) worst planification dimension: lower QoL scores and getting up more than once during the night; (4) worst health dimensions: having a personal chronic disease, lower QoL scores, higher anxiety–mood disorder levels, getting up more than once during the night and age of the PLH individual. No adjustment was performed on the self-esteem score (only one variable with a p value <0.05 in the univariate analysis). The details are described in online supplementary additional table 3

## DISCUSSION

Our study explores, for the first time, the burden of the informal caregivers of PLH individuals whose care is managed at home. Our results are in line with the findings from various countries, reported in the meta-synthesis conducted by Dantas *et al*, showing that parents and families of children with multiple disabilities experience difficulties resulting from the burden of care (health problems, limitations to others activities, increased financial costs).<sup>9</sup> The negative financial impact and the negative planification impact were the most affected dimensions, as previously reported by informal caregivers of patients with other conditions: patients with traumatic brain injury,<sup>19</sup> amyotrophic lateral sclerosis,<sup>20</sup> cancer<sup>21 22</sup> and chronic illness.<sup>21</sup> However, considering that caregiver burden may be defined by positive or negative feelings that may be experienced when giving care,<sup>23</sup> we also found a substantial positive impact on self-esteem. This finding was previously reported in other complex conditions: caregivers of blind children,<sup>24</sup> traumatic brain injury patients,<sup>19</sup> amyotrophic lateral sclerosis patients<sup>20</sup> and cancer patients.<sup>21 22</sup> It is increasingly recognised that the caregiving experience is not only associated with negative consequences, but can result in subjective gains and satisfaction,<sup>25</sup> promote a sense of accomplishment and companionship, and improve self-esteem. The weight of the burden on our sample was very close to those of caregivers of patients with traumatic brain injury,<sup>19</sup> but it was higher than caregivers of cancer patients (Maguire 2016, data not published). We hypothesise that the median age,

**Table 4** Relationships between self-perceived burden of caregivers and their own characteristics

	Self-esteem (0–100)*	Familial support (0–100)†	Financial (0–100)†	Planification (0–100)†	Health (0–100)†
<i>Sociodemographics</i>					
Age					
R	0.076	0.253	0.298*	0.113	0.265*
P value	0.568	0.055	<b>0.022</b>	0.393	<b>0.043</b>
Gender					
Women	80.7±11.6	51.5±17.6	56.1±21.4	68.2±19.1	55.7±20.9
Men	81.5±11.5	55.8±17.9	65.1±21.2	72.1±17.2	53.8±18.1
P value	0.822	0.487	0.103	0.495	0.737
Nature of relationship					
Mothers	81.4±11.1	52.2±17.8	55.2±22.6	70.2±19.4	56.3±21.5
Fathers	81.6±11.7	56.1±18.2	64.5±21.4	73.1±16.8	53.6±18.5
P value	0.958	0.536	0.112	0.710	0.636
Marital status					
Single	83.3±11.0	57.0±12.0	59.3±24.4	72±18.8	59.0±21.0
Not single	80.6±11.6	52.3±18.5	59.3±21.3	69.2±18.4	54.3±19.7
P value	0.486	0.323	0.852	0.530	0.556
Number of children					
R	0.064	0.098	0.144	0.082	0.043
P value	0.605	0.433	0.245	0.511	0.729
Other person with disability at home					
No	81.2±11.6	51.8±17.6	57.5±21.3	68.7±18.4	54.0±20.3
Yes	79.0±11.3	66±14.0	80±13.9	79.3±16.4	65.8±8.01
P value	0.614	<b>0.046</b>	<b>0.012</b>	0.131	0.088
Educational level					
<12 years	82.1±11.1	56.1±16.3	61.8±24.1	70.1±19.7	58.5±22.4
≥12 years	80.2±11.8	50.8±18.5	57.3±19.6	69.2±17.6	52.4±17.5
P value	0.335	0.114	0.347	0.749	0.226
Occupational status					
Worker	79.7±11.4	52.3±20.4	60±20.2	70.2±18.4	56.1±19.2
Not worker	82.4±11.6	53.7±14.8	59.0±23.4	69.7±18.4	54.1±20.9
P value	0.205	0.588	0.843	0.972	0.568
Self-perceived financial status					
Not difficult	80.3±12.2	52.3±18.5	54.0±20.2	67.6±19.5	53.9±20.7
Difficult	82.3±10.2	54.3±16.5	68.9±21.2	73.4±15.7	57.2±18.4
P value	0.425	0.737	<b>0.005</b>	0.209	0.510
<i>Health</i>					
Personal chronic disease(s)					
No	81.7±10.8	52.1±18.4	55.3±22.1	68.8±19.4	51.3±18.8
Yes	79.3±13.1	55.4±15.9	69.2±17.4	71.8±15.7	64.7±19.5
P value	0.549	0.449	<b>0.015</b>	0.705	<b>0.006</b>
Hospitalisation during the last 24 months					
No (55)	81.4±12.2	50.1±16.9	56.7±22.4	67.1±18.3	52.7±19.7
Yes (18)	79.8±9.16	61.7±17.5	67.4±17.2	77.3±17.0	61.9±19.1

Continued

Table 4 Continued

	Self-esteem (0–100)*	Familial support (0–100)†	Financial (0–100)†	Planification (0–100)†	Health (0–100)†
P value	0.383	<b>0.014</b>	0.099	<b>0.021</b>	0.153
<i>Psycho-behavioral characteristics</i>					
Quality of life					
Physical	0.010	−0.471†	−0.554†	−0.465†	−0.544†
P value	0.930	<b>&lt;0.001</b>	<b>&lt;0.001</b>	<b>&lt;0.001</b>	<b>&lt;0.001</b>
Psychological	0.116	−0.421†	−0.354†	−0.352†	−0.405†
P value	0.328	<b>&lt;0.001</b>	<b>0.002</b>	<b>0.002</b>	<b>&lt;0.001</b>
Social	−0.003	−0.468†	−0.336†	−0.441†	−0.425†
P value	0.981	<b>&lt;0.001</b>	<b>0.004</b>	<b>&lt;0.001</b>	<b>&lt;0.001</b>
Environmental	0.132	−0.405†	−0.552†	−0.400†	−0.469†
P value	0.265	<b>&lt;0.001</b>	<b>&lt;0.001</b>	<b>0.001</b>	<b>&lt;0.001</b>
Anxiety–mood					
R	0.061	0.240*	0.165	0.269*	0.378†
P value	0.613	<b>0.043</b>	0.168	<b>0.024</b>	<b>0.001</b>
Coping					
Social support	−0.111	−0.311†	−0.184	0.016	−0.089
P value	0.353	<b>0.009</b>	0.121	0.892	0.457
Problem solvings	0.026	−0.085	−0.160	−0.024	−0.166
P value	0.832	0.487	0.183	0.843	0.166
Avoidance	−0.296*	0.230	0.176	0.237*	0.328†
P value	<b>0.013</b>	0.059	0.144	<b>0.048</b>	<b>0.006</b>
Positive thinking	0.131	−0.235	−0.277*	−0.093	−0.233
P value	0.274	0.052	<b>0.019</b>	0.439	0.051
<i>Burden</i>					
Number of children living at home					
R	0.027	−0.105	0.042	0.007	−0.067
P value	0.837	0.425	0.750	0.956	0.609
Daily caregiving (hours)					
≤4	78.9±11.4	48.5±16.2	54.1±21.0	60±12.0	50±17.9
5–8	82.4±11.5	48.6±17.9	57.1±19.2	66.7±20.7	51.1±17.7
≥9	80.5±11.7	59.5±17.1	64.2±23.9	76.9±16.5	61.2±21.8
P value	0.629	<b>0.037</b>	0.269	<b>0.007</b>	0.092
Get up night (number)					
<2	81.5±10.5	46±15.6	54.5±21.6	61.2±18.0	48.9±19.7
≥2	81.0±12.2	58.7±17.6	63.2±21.4	76.2±15.4	60±18.8
P value	0.982	<b>0.003</b>	0.097	<b>&lt;0.001</b>	<b>0.033</b>
<i>Assistance</i>					
Technical assistance					
No	78.45±9.932	57.66±16.71	66.11±20.13	78±13.95	61.45±21.64
Yes	81.19±12.41	50.05±16.35	58.14±21.88	66.05±18.08	51.42±15.69
P value	0.265	0.060	0.145	<b>0.013</b>	0.072
Human assistance					
No	79.18±12.14	53.75±17.52	62.99±20.99	71.33±18.25	55.83±19.46
Yes	84.61±6.339	50.46±16.29	54.35±21.05	66.46±16.37	54.61±15.20

Continued



**Table 4** Continued

	Self-esteem (0–100)*	Familial support (0–100)†	Financial (0–100)†	Planification (0–100)†	Health (0–100)†
P value	0.133	0.590	0.170	0.368	0.846

R: correlation coefficient

Bold values: p-value&lt;0.05

\*100: best self-esteem.

†100: worse familial support, worse financial impact, worse planification impact, worse health impact.

dependence level and disease evolution of PLH individuals (and, consequently, caregiver burden) are similar to patients with traumatic brain injury, but very different from those of cancer patients who are generally older, less dependent and have less hope of recovery. We found that PLH individuals whose care is managed at their parents' home were young, and presented with a severe and instable PLH with a decompensation of the health status, which may require acute and emergency care. This combination requires the permanent availability of caregivers in a stressful context, which naturally causes burden.

The second part of our findings refers to the factors associated with the burden on informal caregivers. Identifying

burden determinants may help identify unmet needs, prioritise service improvements and support funding decisions. Sociodemographical, health and psycho-behavioural determinants were explored. Contrary to similar studies,<sup>26</sup> our results showed that the main socio-demographical and socioeconomical parameters (such as gender, marital status, educational level and occupational status) were not identified as significant burden determinants. We only found ectopic (and expected) associations. First, we found that health burden was most significant for older caregivers. Complete dependence of the PLH individuals may result in major care and the need to mobilise the patients several times per day (caring,

**Table 5** Relationships between self-perceived burden of caregivers and the characteristics of the polyhandicap child

	Self-esteem (0–100)*	Familial support (0–100)†	Financial (0–100)†	Planification (0–100)†	Health (0–100)†
<b>Age</b>					
R	–0.002	0.263*	0.160	0.099	0.319†
P value	0.984	<b>0.027</b>	0.176	0.406	<b>0.006</b>
<b>Gender</b>					
Boys	79.3±9.98	59.4±17.2	60.1±20.8	73.2±17.6	57.2±20.9
Girls	82.8±12.9	46.5±15.9	58.4±22.8	65.6±18.6	52.6±18.6
P value	0.156	<b>0.002</b>	0.764	0.087	0.441
<b>Severity</b>					
Less severe	79.6±10.9	57.2±17.3	64.4±22.2	74.8±18.0	59.5±18.8
Severe	81.7±11.8	51.0±17.7	56.8±21.1	67.0±18.2	52.8±20.2
P value	0.387	0.171	0.129	0.077	0.164
<b>Stability</b>					
Stable	80.9±12.1	53.2±17.6	59.3±23.0	70.6±19.3	57.4±21.5
Unstable	81.1±10.7	52.8±18.1	59.3±19.9	68.1±17.2	51.5±16.8
P value	0.884	0.829	0.861	0.475	0.249
<b>Medical</b>					
None	80.7±12.1	51.3±16.6	59.4±22.0	68.3±17.7	54.5±19.6
<b>Device</b>					
At least one	82±9.94	57.4±19.9	59±21.2	73.2±20.1	56.5±21.0
P value	0.642	0.246	0.980	0.201	0.825

R: correlation coefficient

Bold values: p-values&lt;0.05

\*100: best self-esteem.

†100: worse familial support, worse financial impact, worse planification impact, worse health impact.



moving, positioning patients and so on).<sup>16</sup> As caregivers age, the burden may increase: families are likely to keep their child at home during the early years, but, as they get older, they are no longer able to take care of them and request the placement of their child in an institution. Some factors modulated burden related to the financial aspects: being older, reports of financial difficulties and caring for a second disabled individual (10% of caregivers cared for a second disabled individual living at home as well as the PLH child). Families were often confronted with reduced financial resources related to work (absence or unstable employment due to time-consuming care) and the necessary expenses for outdoor/indoor facilities or any specialised equipment (eg, wheelchairs). This financial burden deserves the attention of health authorities. While various types of financial assistance in France are proposed to families, they are often confronted with administrative barriers and long lead times. These procedures should be simplified to meet the needs of the families.

As expected,<sup>11 27 28</sup> the caregivers' health status (having a chronic disease, reports of a recent hospitalisation episode, deteriorated emotional status) was related to their reported burden. Preserving caregiver health is a noteworthy issue both for caregivers and, indirectly, for patient health. Health awareness initiative and future health actions should be devised to prevent, diagnose and treat any health (including mental) problems of caregivers.

Psycho-behavioural aspects were strong factors in the modulation of the caregiver burden. First, we found, as reported in previous studies, that QoL, and all its components, was negatively associated with burden. This is supported by previous studies in other contexts, such as caregivers of individuals with mental illness,<sup>29 30</sup> and also caregivers of children with intellectual disability.<sup>28 31</sup> Burden of care and QoL are considered by some authors to be closely related; however, from a theoretical perspective, burden of care and QoL do not measure the same aspects. Caregivers' burden of care is usually defined by its impacts and consequences on caregivers, and there is a negative conceptualisation of caregiving. However, QoL is not a direct conceptualisation of caregiving and proposes a global assessment of the general well-being of individuals.<sup>32 33</sup>

More interestingly and surprisingly, the nature of the coping strategies used played a consequential role in the individual's burden modulation. The association between using avoidance as a coping strategy with worse individual outcomes has been described previously in many various contexts, such as cancer<sup>34</sup> or severe mental illnesses,<sup>35</sup> and also in institutional caregivers of PLH individuals.<sup>36</sup> In contrast, a positive-thinking strategy arose as a protective strategy for burden. A more systematic assessment of coping methods should identify individuals who do not use healthy coping strategies. Targeted psychological interventions based on psychoeducation and cognitive behavioural therapy may be offered to

them.<sup>37 38</sup> Interventions based on problem solving or positive thinking have revealed predictors of satisfactory well-being in caregivers.<sup>39 40</sup>

The objective components of burden were partially related to the subjective self-perceived burden. As previous studies have shown, hours of daily caregiving are associated with greater burden.<sup>7 41</sup> Nocturnal caregiving, which required getting up during the night several times, seemed to be a risk factor for burden. This result suggests the need for reinforcement of human assistance for home care and rest, which consequently may improve caregiver burden. This emphasises the need to maintain specialised institutions allowing alternation between the home and the institution to increase caregivers' free time and respite time while maintaining a family unit and a relationship with the PLH individual. We should be attentive to the various disability policies, according to the principles outlined in the United Nations Convention on the Rights of Individuals with Disabilities ('Disabilities.fr'), which promote a move towards deinstitutionalisation in favour of proximity services, possibly leading to caregiver burden.<sup>9</sup>

Surprisingly, the health status of PLH individuals in terms of severity, stability, medical device and neurodevelopmental status was not associated with the degree of caregiver burden. We can hypothesise that the difficult experience of caring for a PLH child may cause burden and overshadow all other health conditions. However, as the PLH individual aged, the burden level rose, which could be explained by the probable older age of caregivers. An interesting gender difference emerged in the lack of familial support for caregiving: parents of male individuals had less individual support than parents of female individuals confirming our previous results, which demonstrated a lower QoL for the parents of a male child than parents of a female child.<sup>27</sup> This finding referred to the social representations that individuals develop. There is a longstanding and well-documented observation of boy-girl differences in parental affective investments with a male gender 'preference'.<sup>42</sup> Future qualitative studies should further explore these aspects.

### Strengths and limitations

One of the limitations of our study was the small sample size. Our results may benefit from confirmation in a larger sample in the future. However, this is the first study to document the perceived burden on this population of caregivers. This study was based on a cross-sectional design, which did not allow for causality inferences to be made between the studied factors and caregiver burden. Longitudinal designs allow for causality inferences.

### CONCLUSION

This work demonstrated that the caregivers of PLH patients whose care was managed at home experienced a high perceived burden. The main determinants associated with this burden were factors related to the caregivers

themselves (age, health issues and psycho-behavioural aspects), while factors related to the PLH individual were insignificant. Due to the modifiable nature of some of these determinants, detection, prevention and therapeutic strategies should be implemented.

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**Acknowledgements** The French Polyhandicap Group includes the following individuals: Tanguy Leroy, Souhali Haddadou, Cécile Freihuber, Julie Bonheur, Stéphanie Valence, Marie-Christine Nougues, Laurent Luciani, Jean-Pierre Nouët, Catherine Coiffier, Philippe Sellier, Sophie Mathieu, Moustafa Ardati, Delphine Héron, Alexandra Afenjar, Diana Rodriguez, Diana Doummar, Marie-Laure Moutard, Daniel Willocq, Stéphane Pietra, Stéphane Lenormand, Elizabeth Grimont. The authors are grateful to Claire Morando for her logistical support.

**Collaborators** The French Polyhandicap Group includes the following individuals: Tanguy Leroy, Souhali Haddadou, Cécile Freihuber, Julie Bonheur, Stéphanie Valence, Marie-Christine Nougues, Laurent Luciani, Jean-Pierre Nouët, Catherine Coiffier, Philippe Sellier, Sophie Mathieu, Moustafa Ardati, Delphine Héron, Alexandra Afenjar, Diana Rodriguez, Diana Doummar, Marie-Laure Moutard, Daniel Willocq, Stéphane Pietra, Stéphane Lenormand, Elizabeth Grimont.

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**Funding** This work is financially supported by French PREPS (Programme de recherche sur la performance du système de soins, year 2013) and the French Institut National de la Santé et de la Recherche Médicale (INSERM, year 2013): Grant DGOS and INSERM. The sponsor was represented by Assistance Publique, Hôpitaux de Marseille, France; and its role was to control the appropriateness of ethical and legal considerations.

**Competing interests** None declared.

**Patient consent for publication** Not required.

**Ethics approval** Regulatory monitoring was performed according to the French law that requires the approval of the French ethics committee (Comité de Protection des Personnes Sud Méditerranée V, 20/10/2014, reference number 2014-A00953-44).

**Provenance and peer review** Not commissioned; externally peer reviewed.

**Data availability statement** Data are available upon reasonable request.

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