PCR116

Economic Burden and Quality of Life of Caregivers of Patients With Sickle Cell Disease in the United Kingdom and France: A Cross-sectional Study

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INTRODUCTION

- Sickle cell disease (SCD) is a genetic disorder that results in sickle-shaped red blood cells with a reduced ability to deliver oxygen to tissues.[1,2]
- Patients with SCD experience a multitude of symptoms, from acute events (such as painful vaso-occlusive crises and acute chest syndrome) to chronic consequences (such as hemolytic anemia and end-organ damage); both acute and chronic disease sequelae lead to frequent and costly healthcare resource utilization.[1-4]
- Furthermore, children with SCD are at risk of being absent from school, while adult patients and caregivers are often not able to work full-time because of repeated outpatient and inpatient stays.[1,3,5-7]
- Given the high burden of illness and healthcare resource utilization by patients with SCD, the quality of life (QOL) of caregivers of individuals with SCD can be substantially affected by their responsibilities.[5,6,8]

OBJECTIVE

- To capture the impact of caring for individuals with SCD on caregivers' QOL and to estimate the economic burden of SCD using validated questionnaires.

METHODS

- An online survey was developed and administered to caregivers of individuals with SCD in the United Kingdom and France using a market research panel.
- The survey collected sociodemographic data for caregivers and the person they cared for, caregiver QOL data using the Five-level EuroQol 5-dimension questionnaire (EQ-5D-5L)[9,10] and Care-Related Quality of Life instrument with 7 dimensions (CarerQol-7D),[11] and caregiver economic burden data using the Work Productivity and Activity Impairment Questionnaire: Specific Health Problem (WPAI:SHP).[12,13]
  - EQ-5D-5L measured health-related QOL (HRQOL) using the EQ-5D descriptive system, which comprises 5 dimensions: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression.
  - CarerQol-7D measured the impact of providing informal care using both positive and negative dimensions, expressed as "a lot," "some," or "no."
  - WPAI:SHP was adapted to caregivers of patients with SCD to understand the impact of their caring responsibilities on work productivity and regular activities.
- Demographics, HRQOL, and economic burden data were summarized using descriptive statistics.
- EQ-5D-5L scores were converted into utilities by applying the National Institute for Health and Care Excellence (NICE) decision support unit (DSU) value set, which maps EQ-5D-5L data to Three-level EuroQol 5-dimension questionnaire (EQ-5D-3L) utility scores.
  - The French EQ-5D data were analyzed with the French EQ-5D-5L tariff.
  - EQ-5D scores were compared with age- and gender-matched EQ-5D-3L utility scores for the UK participants' age- and gender-adjusted general population norms.
  - Population norms were calculated by using published regression coefficients as utility decrements.
  - Baseline general population health state utility values were generated from the data collected, as they were not available from individuals without caregiver responsibility, to provide a reasonable utility approximation.
- Productivity losses were valued using the human capital method, which values the hourly cost of caring by attaching a market rate value.
- CarerQol-7D utility scores were calculated using the UK tariff value set because there is currently no value set for France.
- Exploratory subgroup analyses were used to calculate the EQ-5D utilities (DSU tariff) and CarerQol-7D results for caregivers with SCD compared with those without SCD; a sensitivity analysis was used to calculate productivity loss using average wage.

RESULTS

Caregiver Sociodemographics

- A total of 69 caregivers, caring for 81 patients with SCD, completed the survey (UK, n=43; France, n=26) (**Table 1**).
- Overall, most caregivers were female, aged 35 to 54 years, and caring for 1 patient with SCD; 22% of caregivers also had SCD (**Table 1**).

Table 1. Sociodemographic Characteristics of Caregivers

Caregiver sociodemographic characteristics	France (n=26)	UK (n=43)	Total (N=69)
Number of patients with SCD under caregiver's care	**n (%)**	**n (%)**	**n (%)**
1	20 (77)	38 (88)	58 (84)
2	5 (19)	5 (12)	10 (14)
3	1 (4)	0 (0)	1 (1)
Age of caregiver			
18-34 years	14 (54)	5 (12)	19 (28)
35-54 years	11 (42)	33 (77)	44 (64)
≥55 years	1 (4)	5 (12)	6 (9)
Sex			
Female	21 (81)	36 (84)	57 (83)
Caregiver has SCD			
Yes	5 (19)	10 (23)	15 (22)
	France (n=33)	**UK (n=48)**	**Total (N=81)**
Relationship	**n (%)**	**n (%)**	**n (%)**
Parent	16 (48)	33 (69)	49 (60)
Child	8 (24)	5 (10)	13 (16)
Other	9 (27)	10 (21)	19 (23)

"Other" relationship category includes aunt/uncle, grandparent, grandchild, partner/spouse, and missing. Percentages for France and the UK are based on the population of respondents within each country. Percentages may not add up to 100 due to rounding.
SCD, sickle cell disease.

EQ-5D-5L

- Using the DSU value set, the mean (SD) caregiver EQ-5D utility scores were 0.66 (0.28) for all respondents, 0.62 (0.29) for the UK sample, and 0.74 (0.34) for the French sample (**Table 2**).
- Using the French value set, the mean (SD) utility scores were 0.78 (0.31) for all responders, 0.74 (0.34) for the UK sample, and 0.84 (0.24) for the French sample (**Table 2**).
- Caregivers of individuals with SCD had a utility decrement of 0.23 when compared with age- and gender-matched population norms (**Table 2**).
- Mean (SD) EQ-5D-5L utility scores for caregivers who had a diagnosis of SCD themselves were lower than those of caregivers without SCD: 0.62 (0.23) vs 0.67 (0.53), respectively (**Table 2**).
- The dimension with greatest effect on caregivers (defined as the dimension with the greatest amount of "extreme" and "severe" problems) was pain/discomfort; the least affected dimension was self-care.

Table 2. Caregiver EQ-5D Utility Scores[a]

EQ-5D results	UK (DSU) tariff Mean (SD)	French tariff Mean (SD)
Pooled sample, N=69	0.66 (0.28)	0.78 (0.31)
UK sample only, n=43	0.62 (0.29)	0.74 (0.34)
French sample only, n=26	0.74 (0.34)	0.84 (0.24)
Age- and gender-matched EQ-5D utility difference	**Decrement[b]**	
UK (DSU) tariff, pooled sample, N=69	0.23	
Subgroup analysis	**Mean (SD)**	
Caregiver has SCD, n=15	0.62 (0.23)	
Caregiver does not have SCD, n=52	0.67 (0.53)	

[a]EQ-5D utility scores range from 0 (a state as bad as being dead) to 1 (full health).
[b]The decrement between general population age- and gender-matched utility score for the pooled overall sample was calculated as follows: population age- and gender-matched utility (UK 0.888) – SCD sample utility (UK 0.661) = 0.227.
DSU, decision support unit; EQ-5D, EuroQol 5-dimension questionnaire; SCD, sickle cell disease.

CarerQol-7D

- The mean (SD) caregiver CarerQol-7D utility score was 80.69 (24.40) (**Table 3**).
- The CarerQol-7D utility score for caregivers who had a diagnosis of SCD was 71.87, compared with 83.90 for those without SCD (**Table 3**).

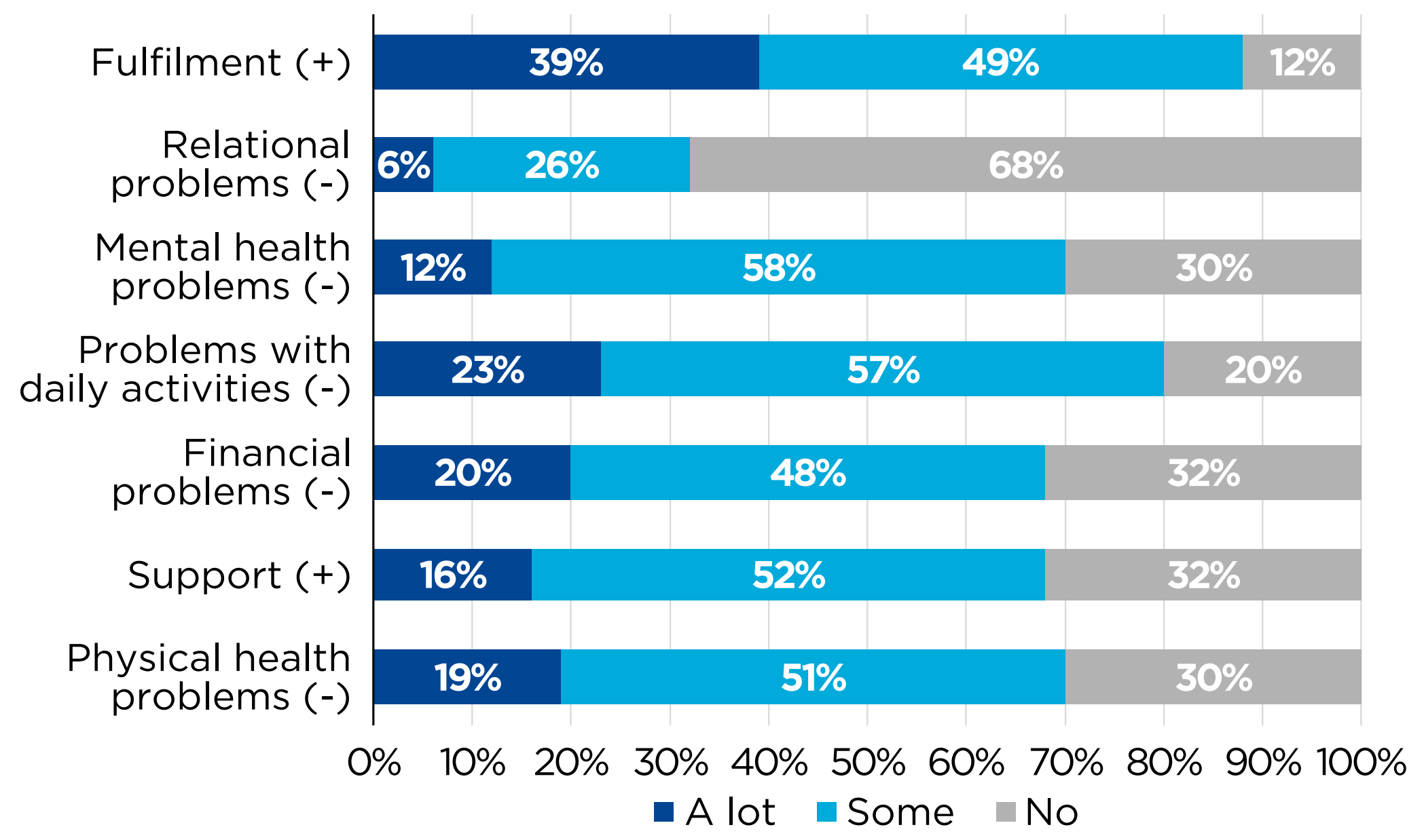
Table 3. Caregiver CarerQol-7D Utility Scores[a]

CarerQol-7D	Mean (SD)
Pooled sample, N=69	80.69 (24.40)
UK sample only, n=43	78.72 (25.79)
French sample only, n=26	83.97 (22.01)
Subgroup analysis	**Mean (SD)**
CarerQol-7D caregiver has SCD	71.87 (23.13)
CarerQol-7D caregiver does not have SCD	83.90 (24.40)

[a]CarerQol-7D utility scores range from 0 (worst imaginable caregiving situation) and 100 (best imaginable caregiving situation).
CarerQol-7D, Care-Related Quality of Life instrument with 7 dimensions; SCD, sickle cell disease.

- The most affected dimension was caregivers' problems with daily activities, and the least affected dimension was caregivers' relational problems with the individual(s) they care for (**Figure 1**).
- Sixty-eight percent of all caregivers indicated issues in the financial problems dimension (**Figure 1**).
- Eighty-eight percent of caregivers expressed "a lot" or "some" in the fulfilment dimension (**Figure 1**).

Figure 1. CarerQol-7D Dimensions (Total Sample)



(+) represents the positive aspects of caring ("a lot" = best level); (–) indicates the negative aspects of caring ("a lot" = worst level).
Permission to translate the CarerQol-7D into French using a forward and backward translation method by translators who have International Organization for Standardization certification was obtained from the instrument developers.
CarerQol-7D, Care-Related Quality of Life instrument with 7 dimensions.

WPAI

- Overall, 37 caregivers were employed and completed work-related WPAI questions.
- The mean (SD) percentages of impairment due to caregiving (n=37) were
  - Absenteeism 22% (24%).
  - Presenteeism 46% (24%).
  - Overall work impairment 34% (19%).
- Among all caregivers (N=69), the mean (SD) for activity impairment was 53% (24%).
- The mean weekly value of productivity loss per caregiver was £87.68 in the UK and €72.61 in France, resulting in an average yearly loss of £4208 and €3485 when calculated as the 2022 minimum wage (£9.50 and €10.85 in the UK and France, respectively), rising to £5391 and €9319 when using the 2022 average wage rate (£12.17 and €29.01) (**Table 4**).

Table 4. Caregiver Productivity Losses

Base case[a]	Monthly mean economic loss	Annual mean economic loss
UK, n=24	£351	£4209
France, n=13	€290	€3485
Sensitivity analysis[b]	**Monthly mean economic loss**	**Annual mean economic loss**
UK, n=24	£449	£5391
France, n=13	€777	€9319

The "n" values represent an employed subsample.
[a]Base case analysis – monetary value as the minimum wage. [b]Sensitivity analysis – monetary value using average wage rate.

CONCLUSIONS

- To our knowledge, this is the first study that has measured HRQOL using the EQ-5D-5L and the CarerQol-7D in caregivers who provide care for individuals with SCD in the United Kingdom and in France.
- The calculated age- and gender-matched utility decrement of 0.23 compared with population norms demonstrates that caring for individuals with SCD affects many aspects of caregivers' HRQOL.
- Notably, the CarerQol-7D utility score in this study (80.69) was lower than CarerQol-7D scores reported for other rare diseases, eg, cystic fibrosis (84.6).[14]
- This study demonstrates that along with high levels of productivity and economic losses, caregiver mental health is particularly impacted.
- These findings, which highlight both the economic and psychosocial burden of caregiving for patients with SCD, may be useful for Health Technology Assessment submissions not only to ensure access to effective SCD treatment but also to alleviate the impact of caring for patients with SCD.

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DISCLOSURES

- In accordance with good practice, both the information sheet and the main survey were reviewed by an independent ethics reviewer working under the auspices of the Association of Research Managers and Administrators.
- **Martin Besser:** honoraria: Sanofi, Novartis; advisory boards: Novartis, Global Blood Therapeutics, Forma, Amgen, Hemeo; educational support: Grifols, Novartis, Global Blood Therapeutics. **Ewa Drogon O'Flaherty:** employee, equity ownership: Global Blood Therapeutics. **Sian Bissell O'Sullivan:** employee: PHMR. **Siobhan Bourke:** employee: PHMR. **Yemi Oluboyede:** employee: PHMR. **Giovanna Barcelos:** employee: ProClinical AG; employee, equity ownership: Global Blood Therapeutics. **Matthew Binns:** employee, equity ownership: Global Blood Therapeutics. **Louise Longworth:** employee: PHMR.
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