Bearing the Brunt: Disproportionate Burden Among Female Parent-Caregivers in Rare Disease

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Background

- Caring for a child with a rare disease takes a heavy toll on parents, resulting in poor mental and physical health, reduced quality of life (QoL), and a substantial economic burden.[1-3]
- Much of the literature on caregiver burden emphasizes the role of primary caregivers. While mothers are broadly reported as the primary caregiver, most studies do not distinguish maternal and paternal experiences.

Objectives

- The objective of this research was to differentiate maternal and paternal caregiving burden in pediatric rare diseases.

Methods

- We conducted a targeted literature review in PubMed, using search terms for rare diseases, parental caregiving, and caregiver health, QoL, and economic outcomes.
- A citation-mining approach identified additional articles from the reference lists of key publications and citations, focusing on English-language papers published from 2018 to 2023.
- Articles selected for inclusion compared one or more outcomes between female and male parent caregivers.

Results

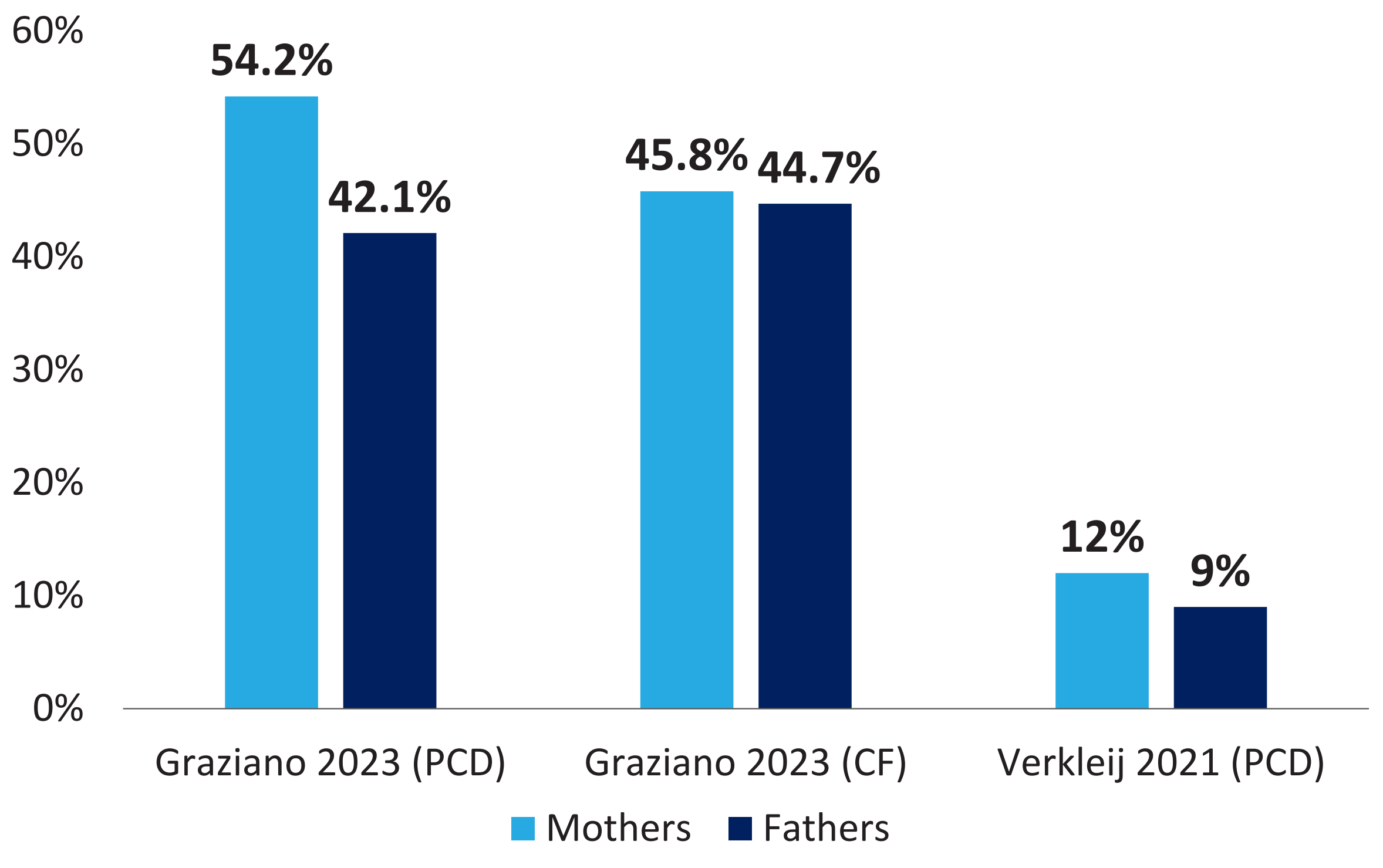
Included studies

- The 11 identified studies covered cystic fibrosis (CF), cancer, Dravet syndrome, primary ciliary dyskinesia (PCD), and tuberous sclerosis complex.

Caregiver mental health

- Several studies found higher rates of depression and anxiety among female versus male parent caregivers, as shown in **Figures 1 and 2.**[4-6] Furthermore, in one study, suicidal ideation was reported in 20% of mothers of children with PCD, compared with 10% of fathers.[4]
- While both mothers and fathers of children with cancer had an increased rate of psychiatric hospital visits relative to matched controls, the incremental risk was consistently greater over time among mothers versus fathers.[7]

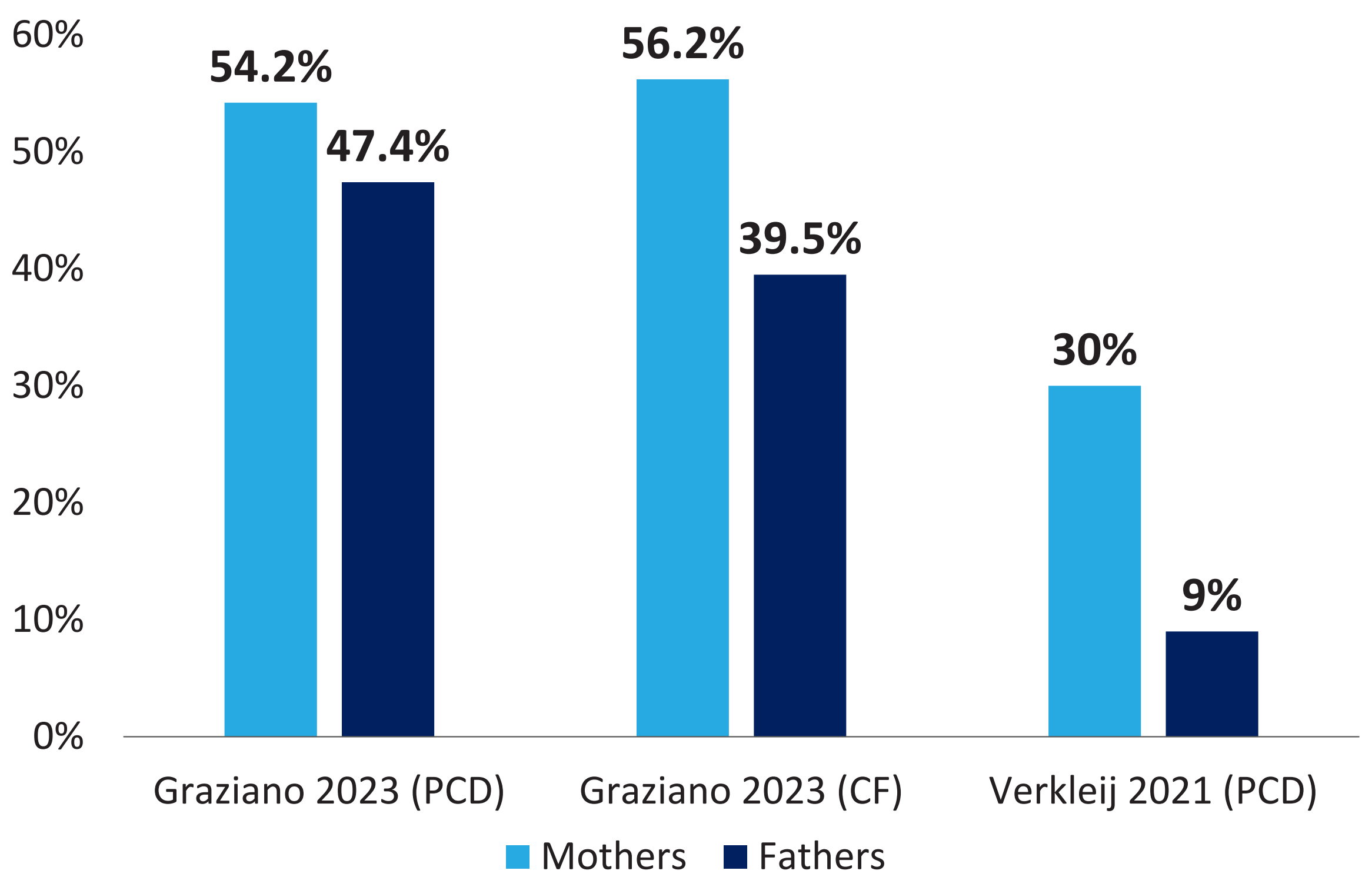
Figure 1. Clinically elevated depression among mothers and fathers[4,5]



CF = cystic fibrosis; PCD = primary ciliary dyskinesia

Results

Figure 2. Clinically elevated anxiety among mothers and fathers[4,5]



CF = cystic fibrosis; PCD = primary ciliary dyskinesia

Caregiver quality of life

- One study of parents of children with CF found being a mother to be independently associated with high caregiver burden (odds ratio [OR]: 1.65; 95% CI: 1.02–2.65; p<0.05).[8]
- Among parents of children with tuberous sclerosis complex, significantly lower QoL was observed among mothers versus fathers, as measured by the EQ-5D (time-trade off: p=0.020; visual analog scale [VAS]: p=0.039).[6]
- Higher median utility scores demonstrated a significantly lower burden for fathers (89.2 [interquartile range (IQR) 79.6–96.5]) compared to mothers (84.7 [IQR: 74.5–88.0]); p<0.001) of children with CF.[8]
- A study of parents of children with CF and PCD found a higher caregiver burden among mothers versus fathers, although the difference was not significant.[9]
- Of five studies identified in a systematic literature review analyzing the impact of gender on QoL among parents of children with rare diseases, one study reported lower psychosocial QoL in mothers versus fathers, while four studies found no significant gender differences.[10]

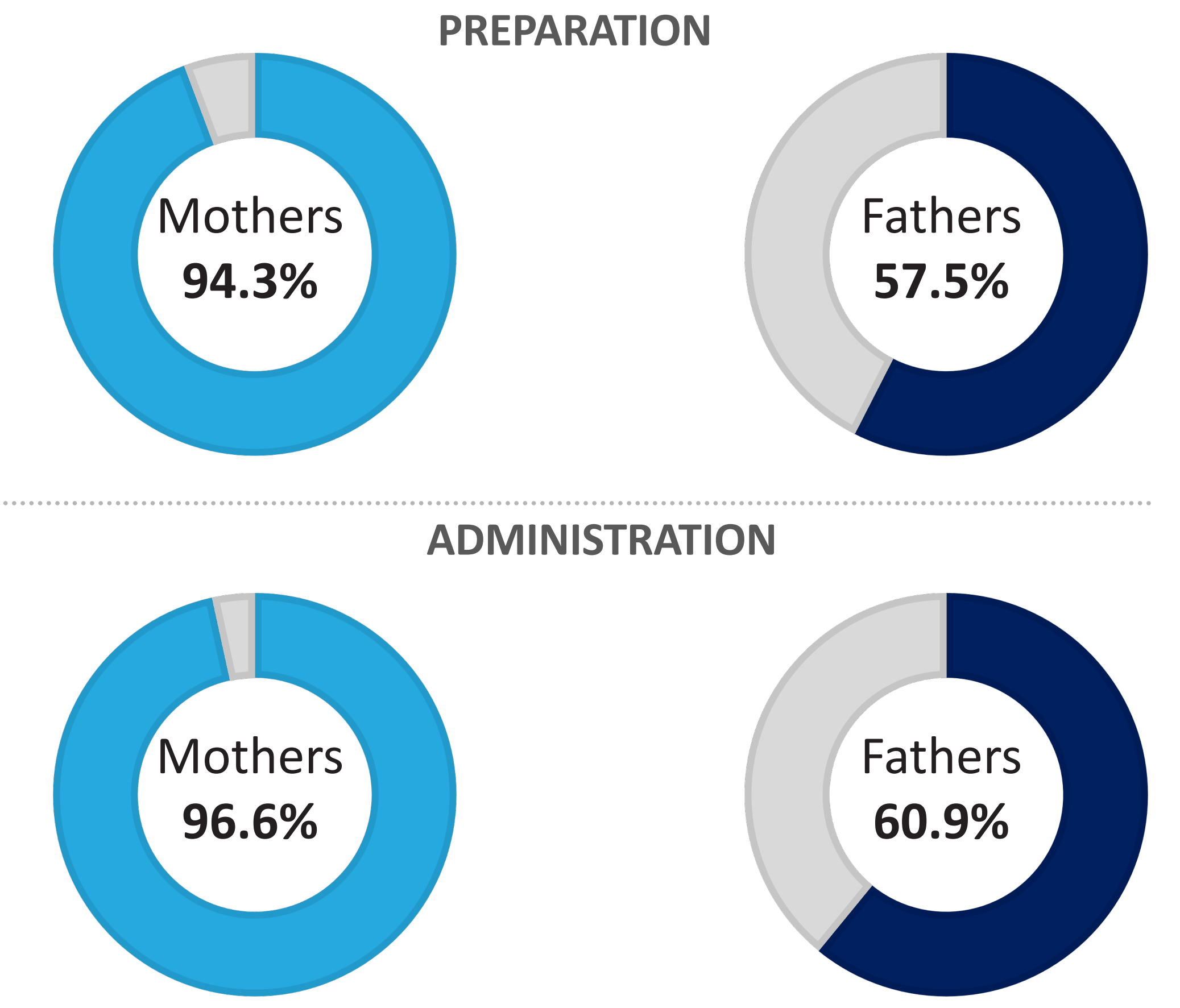
5.8x greater care burden

Female caregivers of children with cancer experienced 5.8 times the caregiver burden of male caregivers (p=0.004).[11]

- Notably, in a study of children with CF and their parents, the mothers' caregiver burden was associated with the QoL of the child to a greater extent than was the fathers' burden.[9]
  - Maternal burden demonstrated a significant inverse correlation with Cystic Fibrosis Quality of Life, Revised (CFQOL-R) physical and emotional subscales, Cystic Fibrosis Questionnaire, Revised (CFQ-R) body image, CFQ-R treatment burden, CFQ-R eating, and CFQ-R gastrointestinal assessments.[9]
  - Paternal burden demonstrated a significant inverse correlation with CFQ-R emotional and CFQ-R respiratory assessments.[9]

- A study of children with Dravet syndrome and their parents found that more mothers than fathers participated in treatment administration and preparation for ~30 minutes each day (**Figure 3**), representing a substantial use of time that may influence QoL.[12]

Figure 3. Participation in daily treatment preparation and administration[12]

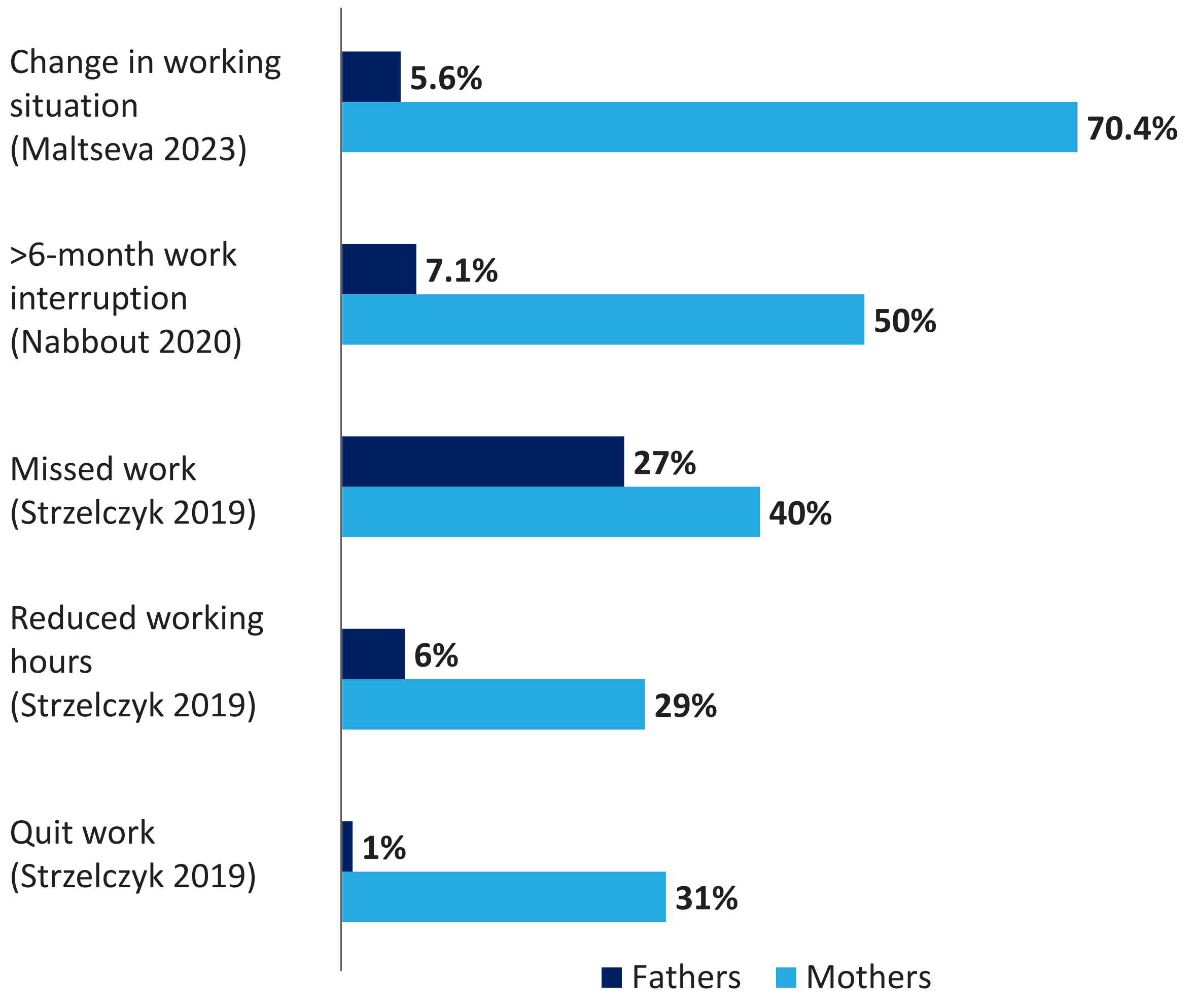


- Having a child with Dravet syndrome impacted most parents' social lives; just 3.4% of mothers and 8.3% of fathers reported no impact.[12]
- More mothers than fathers reported caregiving having an important impact on their time and energy (85.1% versus 49.4%).[12]

Productivity and economic impacts

- In three studies of parent caregivers of children with Dravet syndrome, impacts to work and productivity were substantially greater among mothers compared with fathers (**Figure 4**).[12-14]

Figure 4. Impacts to work among mothers and fathers of children with Dravet syndrome[12-14]



- Annual indirect costs resulting from caregiving-related lost productivity were more than 10 times greater among mothers versus fathers in a study of children with Dravet syndrome (**Table 1**).[14]

Table 1. Indirect costs associated with Dravet syndrome among mothers and fathers[14]

	Mean costs over 3 months	Mean annual costs
Maternal indirect costs	**€ 4,399**	**€ 17,594**
Quit work	€ 3,170	€ 12,679
Reduced working hours	€ 732	€ 2,930
Missed days of work	€ 496	€ 1,986
Paternal indirect costs	**€ 391**	**€ 1,565**
Quit work	€ 109	€ 437
Reduced working hours	€ 155	€ 618
Missed days of work	€ 127	€ 509

Discussion

- This literature review found a substantial burden associated with caregiving among parents of children with rare diseases, including impacts to mental health, QoL, productivity, and work.
- While this burden exists among males and females, it is particularly pronounced among female parent caregivers.
- Understanding the factors associated with increased caregiver burden can contribute to prevention initiatives and help inform efforts to alleviate burden among parents of chronically ill children.
- In some familial situations, it may be beneficial to identify barriers to more equitable distribution of caregiving responsibilities, thus helping to reduce the burden on female caregivers.
- Further study is needed to assess social and cultural influences such as gender differences in reporting mental health concerns and accessing care, as well as the influence of gender roles in work and family responsibilities.

Conclusions

- The burden associated with caregiving among parents of children with rare disease appears to disproportionately affect females.
- Identifying female gender as a risk factor for caregiver strain is key to prevention and early identification of the mental health, QoL, and economic impacts experienced by this population.

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