

Comparing the humanistic and economic impact of osteogenesis imperfecta affected and non-affected caregivers, who care for an individual with osteogenesis imperfecta

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Background

- Osteogenesis imperfecta (OI) is a rare, heritable connective tissue disorder with multiple manifestations, and variable severity. Individuals with OI typically have low bone mass and skeletal fragility, and are susceptible to morphometric vertebral fractures and compression, bone deformities, scoliosis, and growth deficiency.¹
- The IMPACT Survey aimed to describe the clinical, humanistic and economic impact of OI on the OI community.²
- This analysis quantifies the impact of caring for an individual with OI whilst also having OI on quality of life (QoL), finances and productivity and compares the impact to caregivers (CG) of individuals with OI, who do not have OI themselves, referred to as CGs with and without OI respectively.³

Results

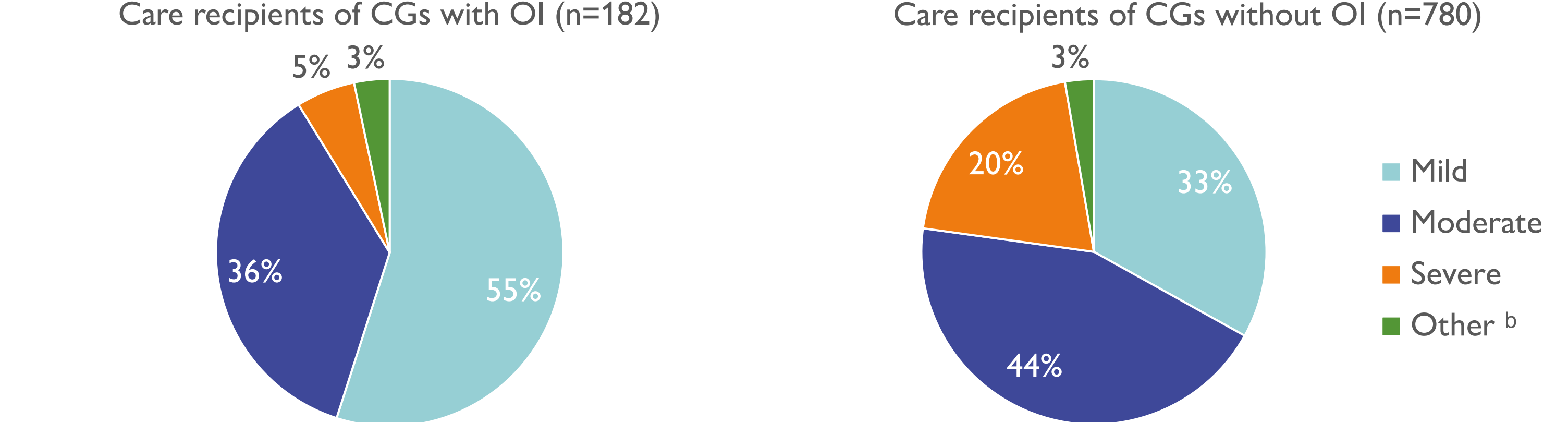
Demographics and respondent characteristics

- A total of 150 CGs with OI and 560 CGs without OI responded to the survey.
- The mean age of CGs with and without OI was 42 years (ranges: CGs with OI, 24–75; CGs without OI, 18–81).
- Most CGs were female (CGs with OI 72% female; CGs without OI 83% female).
- CGs with OI reported mostly mild OI severity for care recipients. Those without OI mostly reported moderate severity (Figure 1).
- Caring for multiple care recipients with OI was more common for CGs with OI vs those without (>1 care recipient 25% vs 6%, respectively)
- CGs with OI were in similar levels of employment (64% employed) to CGs without OI (65% employed).

Methods

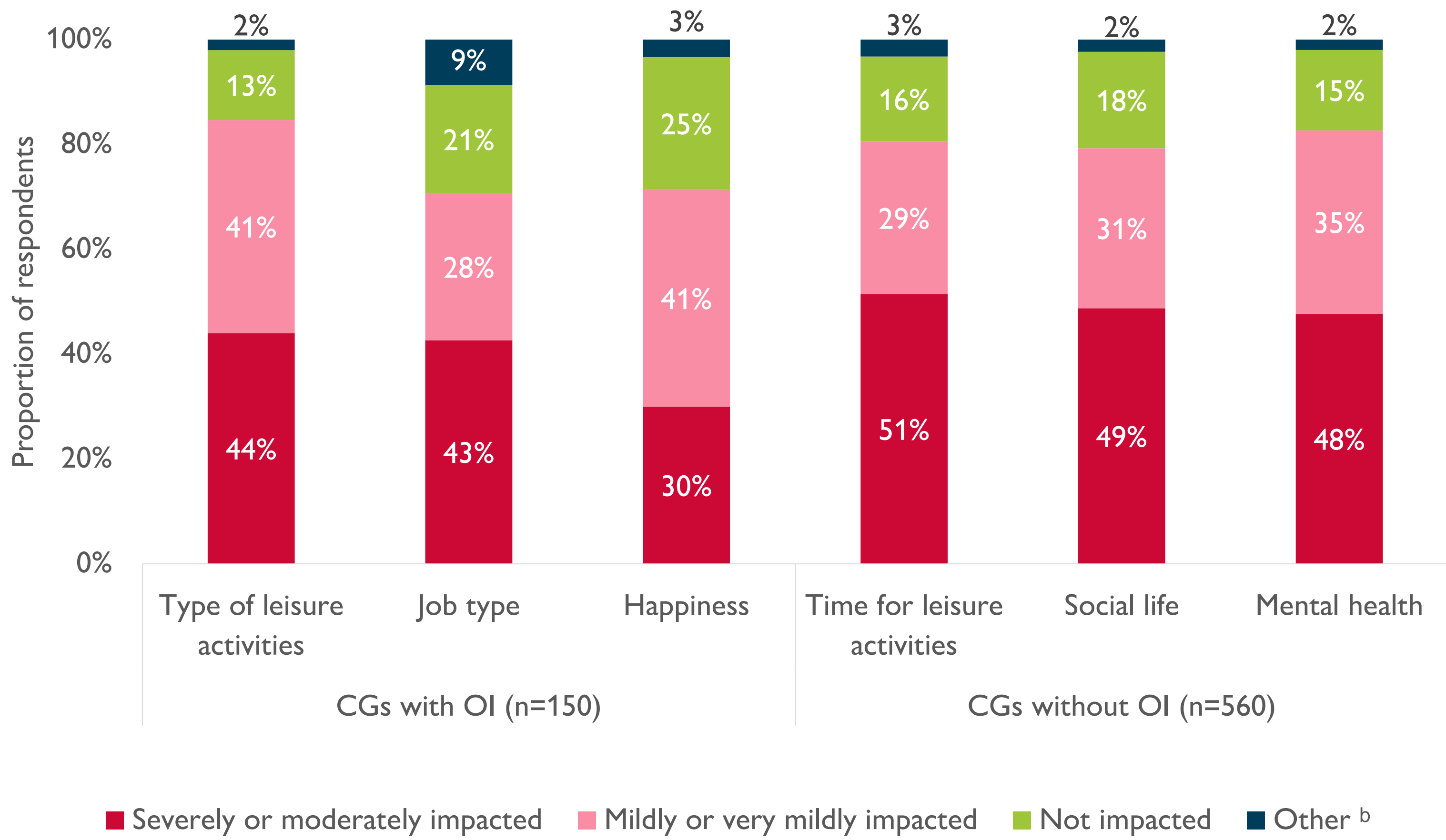
- Developed by the Osteogenesis Imperfecta Federation Europe (OIFE), the Osteogenesis Imperfecta Foundation (OIF) and an international steering committee of OI clinical experts.
- Aimed at adults with OI, CG (with or without OI) of children or adults with OI, adolescents with OI and close relatives.
- Included up to 102 questions on the clinical, economic, and humanistic impact of OI.
- Was professionally translated into eight languages and fielded online July–September 2021.
- Access to IMPACT Survey data available on request at www.impactsurveyoi.com.
- Microsoft Excel was used to clean, code, and analyse data.

Figure 1. CG assessed OI severity of care recipients (n=962)



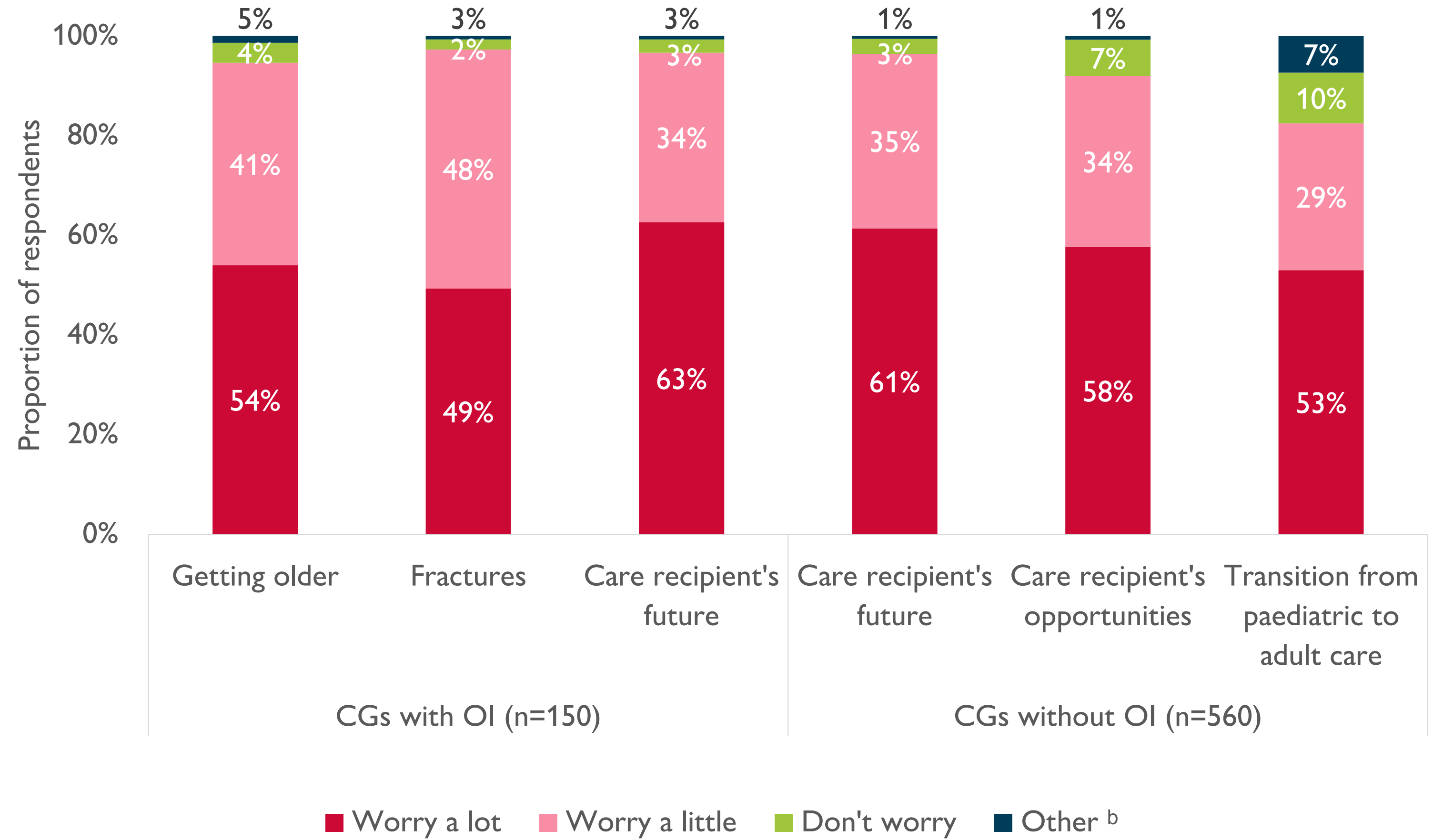
Humanistic impact of caregiving

Figure 2. Most impacted areas of QoL due to OI (n=710)



- CGs with and without OI were both asked questions about their careers, social life, relationships and mental and physical health. CGs with OI were also asked questions on their ability to live independently and selfcare.
- Among CGs with OI any impact was most commonly reported on type of leisure activities (85%), job type (71%) and happiness (71%); for CGs without OI these were time for leisure activities (80%), social life (80%) and mental health (83%).

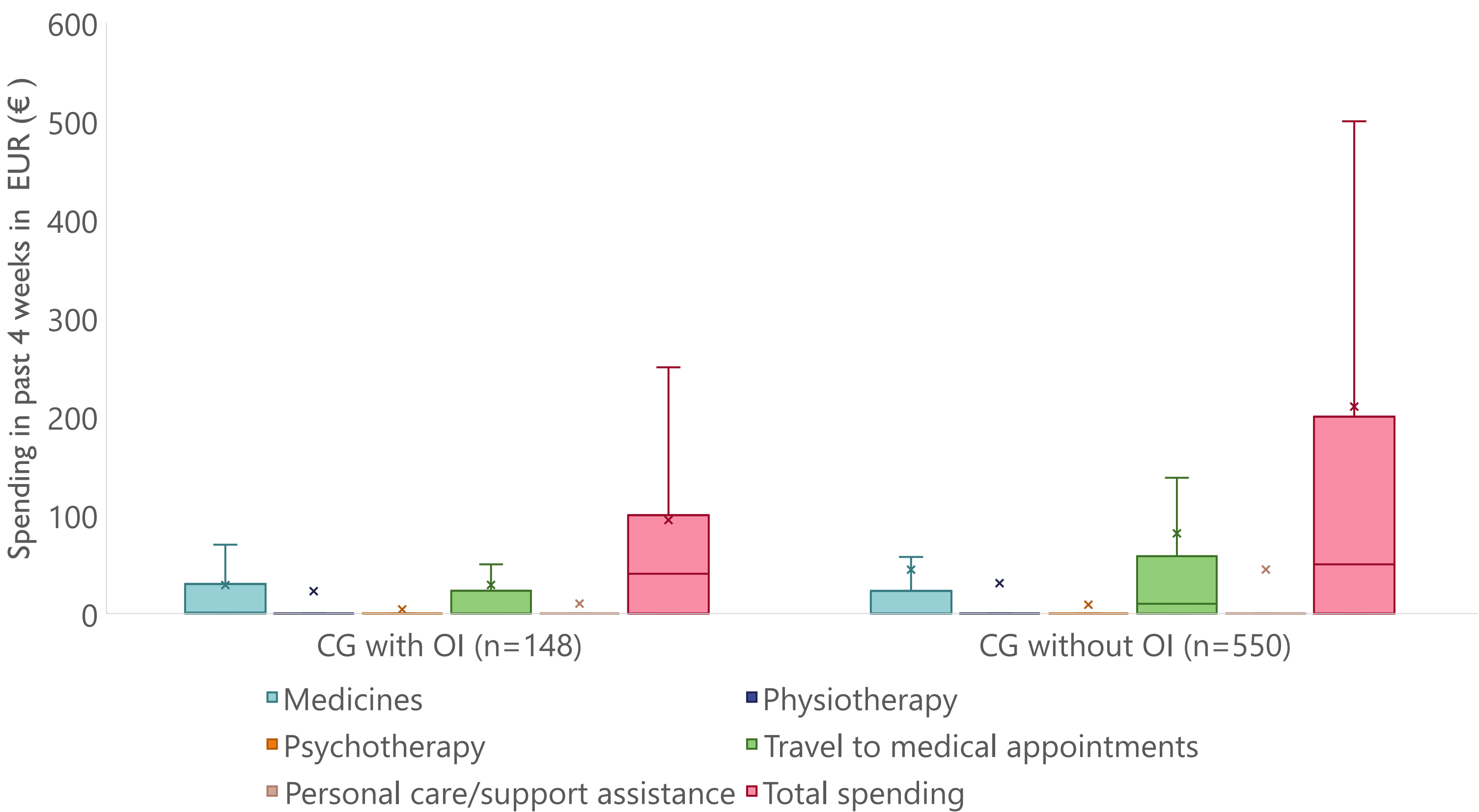
Figure 3. Areas where highest proportions of caregivers worried (n=710)



- CGs with and without OI were both asked questions about access to care, side effects, relationships, finances and care recipients. CGs with OI were also asked questions about their own OI.
- The most common areas of worry (either a lot or a little) for CGs with OI were getting older (95%), fractures (97%) and care recipient's future (97%); for CGs without OI these were their care recipient's future (96%), opportunities (92%) and transition from paediatric to adult care (82%).

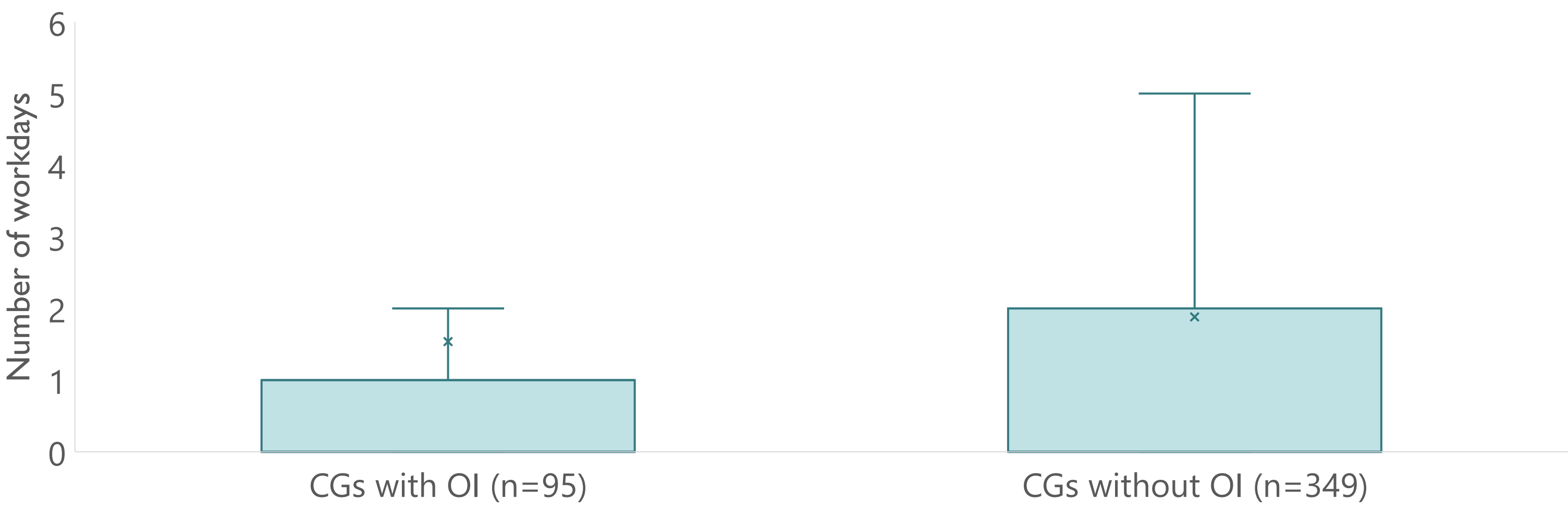
Economic impact of caregiving

Figure 4. OI-related out-of-pocket expenses in a four-week period (n=698)^{c,d}



- CGs without OI had a higher total spend with larger variability (mean €210, standard deviation [SD] €658), compared with CGs with OI (who reported spending on themselves and care recipients; mean €95, SD €174).
- For CGs with OI spending on medicines was highest mean (€29, SD €74); for those without OI, most was spent on travel to medical appointments (mean €81, SD €343).

Figure 5. Missed workdays due to OI in a four-week period (n=444)^e



- In a four-week period, CGs without OI missed slightly more workdays compared with those with OI (mean 1.9 days SD 3.9 vs 1.5 days, SD 4.6).
- The distribution of CGs in full-time, part-time and self-employment were similar across both groups.
- Both groups missed more workdays than the UK general population (0.35 days in 4 weeks).²

Conclusions

- CGs experience a considerable impact on their QoL, productivity, and finances, whether they have OI or not.
- Limited differences in the humanistic impact for CGs with and without OI were identified. The full impact on CGs with OI may be hidden by the effect of the disability paradox, which may make caregivers with OI more resilient to negative impact.³
- Comparison of the economic impact on CGs with and without OI is challenging due to differences in sample sizes, OI severity of care recipients and the number of care recipients.

Acknowledgments

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References and footnotes

References: [1] Marini J.C., et al. 2017. Nat Rev Dis Primers;3:17052 [2] Hart T, et al. 2024. Orphanet J. Rare Dis; 19:222. [3] Albrecht G, et al. 1999. Soc Sci Med; 48(8):977-88.

Footnotes: ^a Question numbers analysed were 1,8,18,22,46,55, 80–83, 86, 92, 102–105, 162 and 170; ^b Includes the answer options “Other”, “Prefer not to say” and “I don’t know”; ^c Out-of-pocket costs were converted into Euros (€) using the conversion rate in effect on July 1, 2021; ^d Respondents who indicated their use of Chilean peso were excluded due to complexities arising from the unusual currency conversion rate; ^e Only asked to respondents in paid employment