

A global perspective on self-reported productivity losses associated with osteogenesis imperfecta

Ingunn Westerheim¹, Taco van Welzenis¹, Lena Lande Wekre², Cathleen Raggio³, Michael B Bober⁴, Oliver Semler⁵, Tracy Hart⁶, Ruby Dadzie⁷, Maria Rapoport⁷, Samantha Prince⁷, and Frank Rauch⁸

¹ Osteogenesis Imperfecta Federation Europe, Heffen, Belgium; ² Sunnaas Rehabilitation Hospital, Nesoddtangen, Norway; ³ Hospital for Special Surgery, New York, USA; ⁴ Alfred I. duPont Hospital, Wilmington, Delaware, USA; ⁵ University of Cologne, Germany; ⁶ Osteogenesis Imperfecta Foundation, Gaithersburg, Maryland, USA; ⁷ Wickenstones Ltd, Abingdon, Oxfordshire, UK; ⁸ McGill University, Montreal, Canada.

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 deficiency¹
- The IMPACT Survey explored the clinical, humanistic and economic impact of OI on people with OI, their caregivers (CG) and wider society
- Here, we present the global findings on OI-associated productivity losses for non-CG adults with OI, and CG with and without OI

Methods

The IMPACT Survey

- Developed by the Osteogenesis Imperfecta Federation Europe (OIFE), the Osteogenesis Imperfecta Foundation (OIF) and an international steering committee of OI clinical experts

Results

Demographics and respondent characteristics

- Overall, 2,208 participants were recruited across 66 countries
- Of 2,000 adult respondents, 1,290 were non-CG with OI, 150 were CG with OI, and 560 were CG without OI (Figure 1)
- Women were more highly represented than men (70% of adults with OI were female, 83% CG without OI were female)
- Most respondents with OI reported moderate OI severity (47%), over a third reported mild (35%), and fewer reported severe OI (14%)
- The mean age of non-CG with OI was 43 years (range 18–85 years), CG with OI 42 years (range 24–75 years), and CG without OI 42 years (range 18–81 years)

Figure 2. Employment status by respondent category

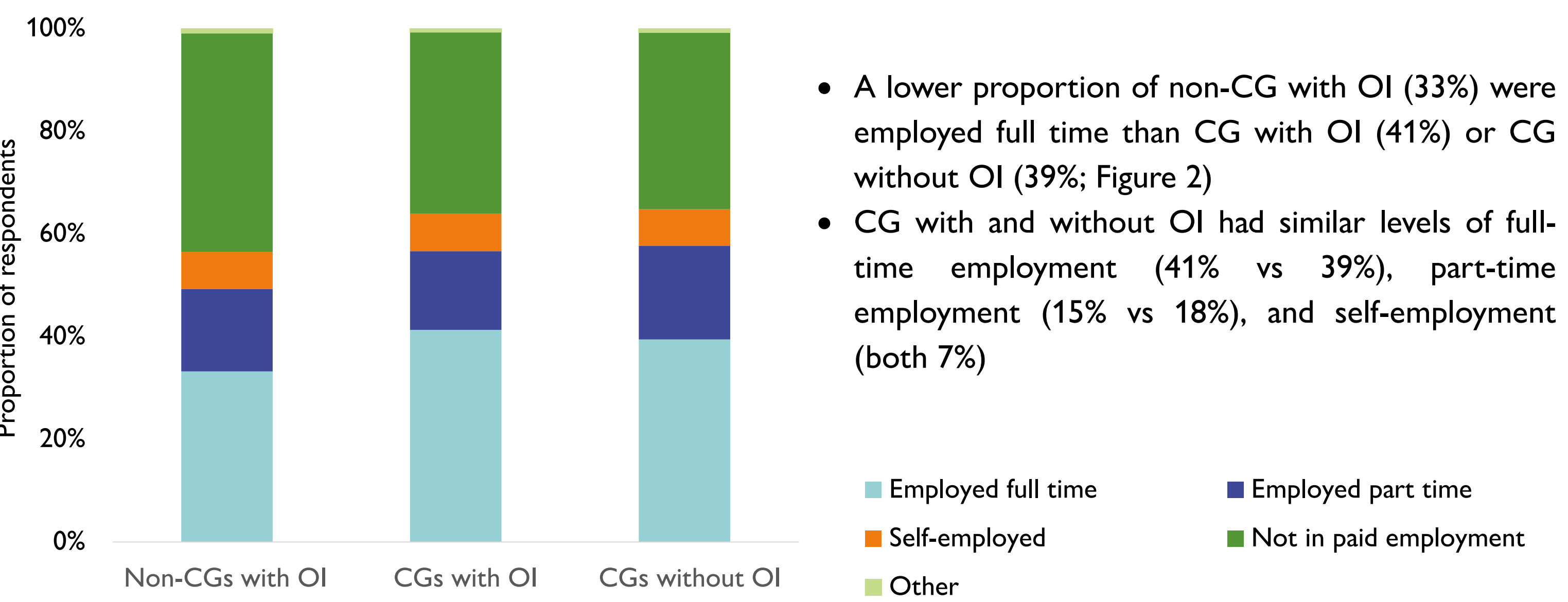


Figure 4. Occupation other than paid employment by respondent category

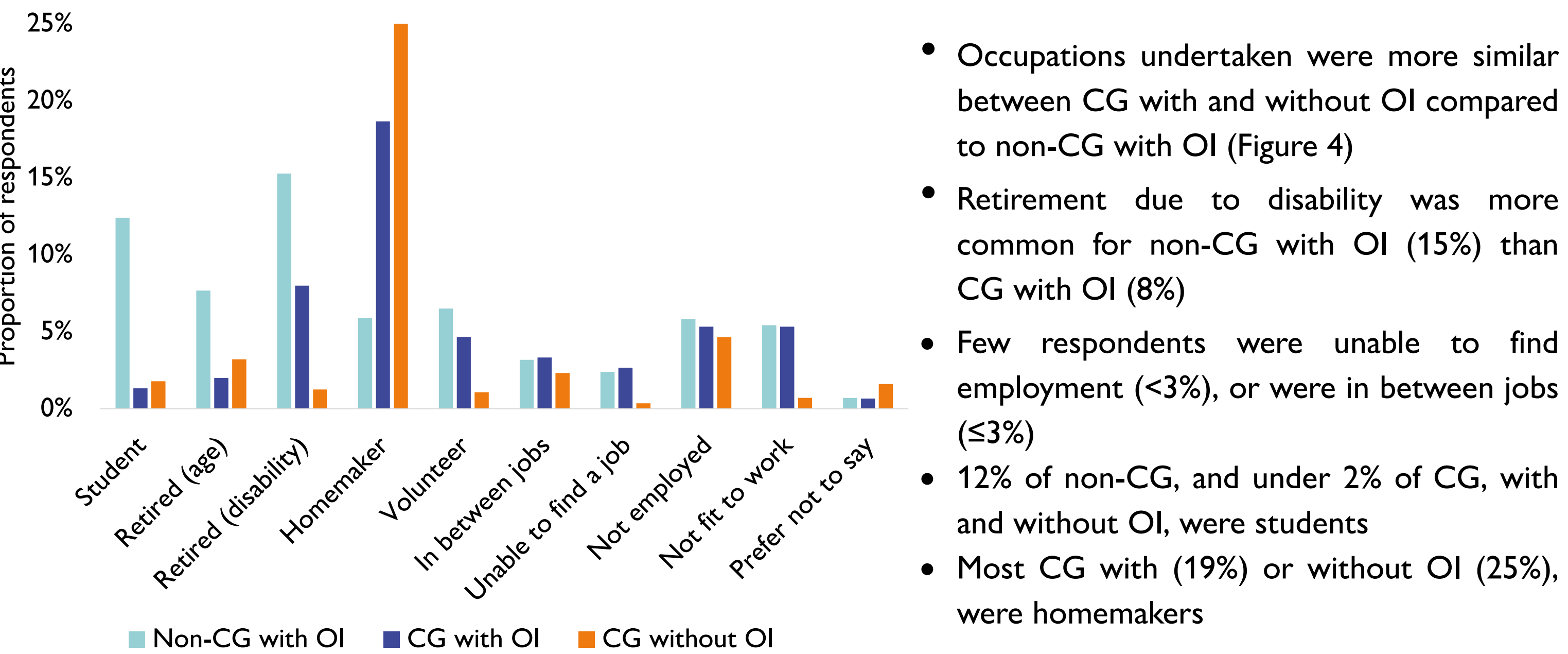
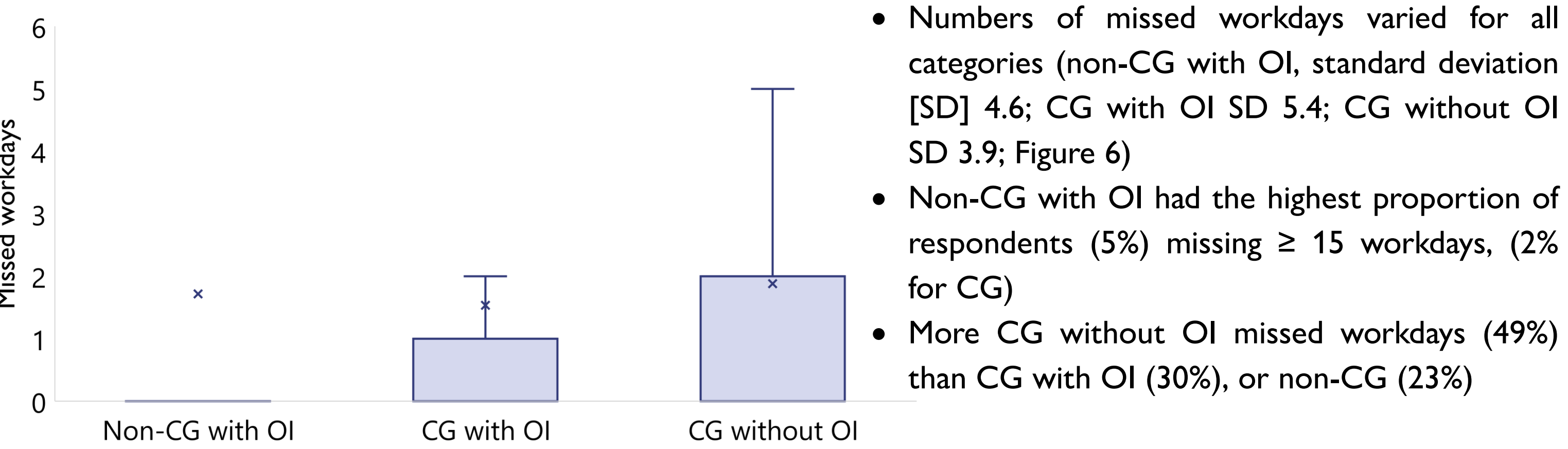


Figure 6. Missed workdays in a 4-week period by respondent category



Childcare (excluding school, and medical care) for children with OI (CG without OI)

“When small, [I] paid private sitter due to daycares refusing to care for [my child] due to OI”
- USA

“Siblings are doing things they shouldn’t need to do out of love”
- Sweden

- Most CG without OI (82%) provided support for their children, often alongside their partners (73%; data not shown)
- Just over half of respondents received support from other relatives (56%)
- 11% of respondents paid for support, and a minority received free, professional support (social services, 6%; charities, 1%)

Methods (continued)

- Aimed at adults with OI, CG (with or without OI) of children or adults with OI, adolescents with OI and relatives
- Included up to 102 questions on the clinical, economic and humanistic impact of OI
- Was professionally translated from English into French, European Spanish, Latin American Spanish, Portuguese, Russian, German, Italian, and Dutch, and fielded online July–September 2021

Recruitment

- Advertised through emails, meetings and social media engagement by the OIFE, and OIF

Analysis

- Responses from all languages were professionally translated into English
- Microsoft Excel was used to clean, code, and analyse data

Figure 1. Number of adult respondents by continent

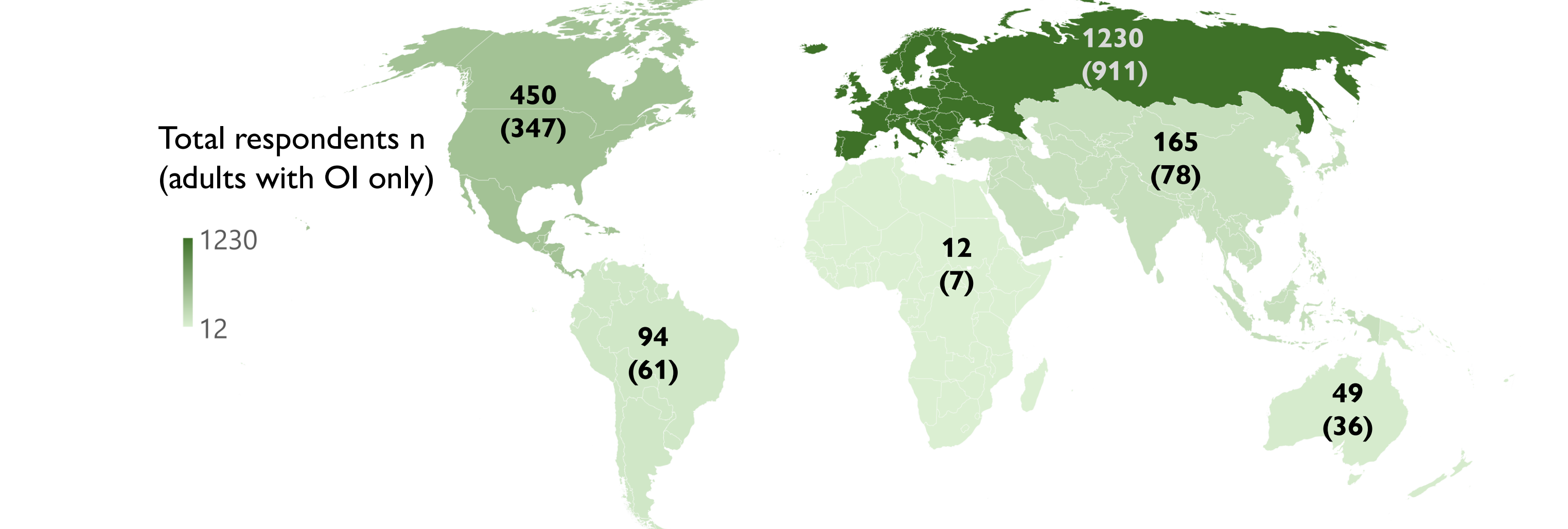


Figure 3. Employment status by OI severity (adults with OI)

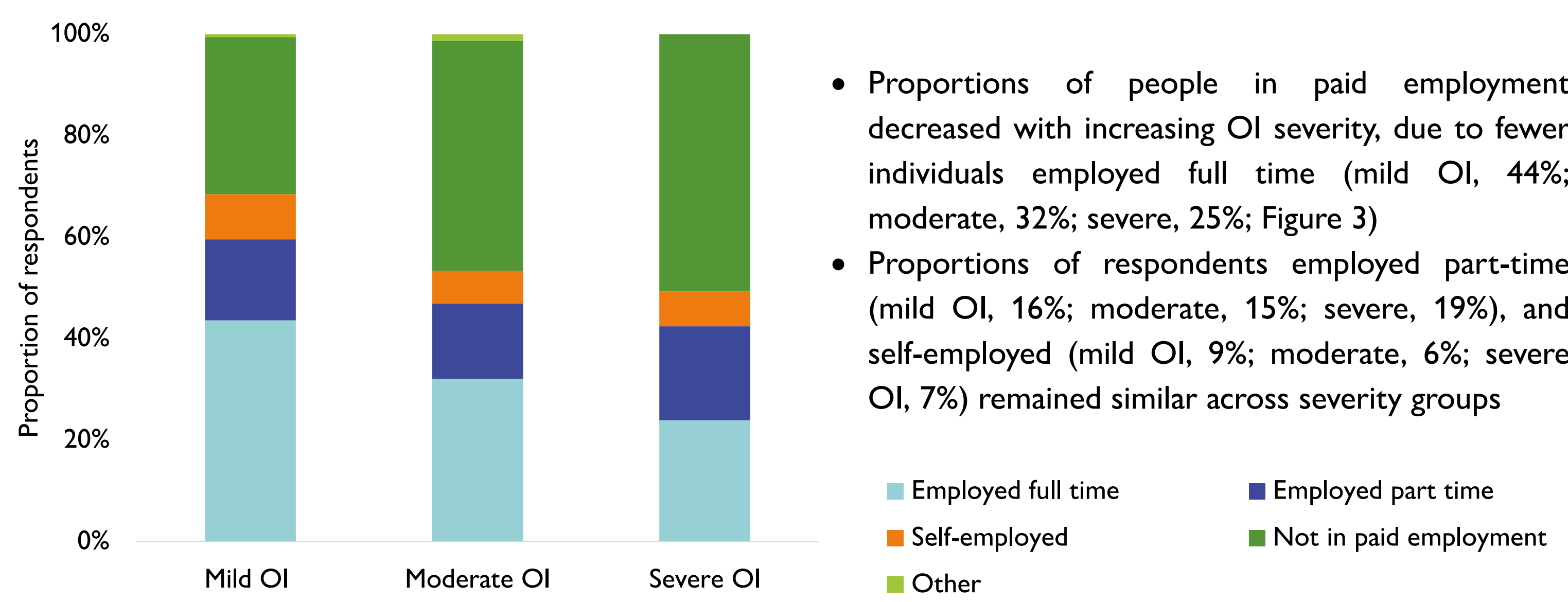


Figure 5. Occupation other than paid employment by OI severity (adults with OI)

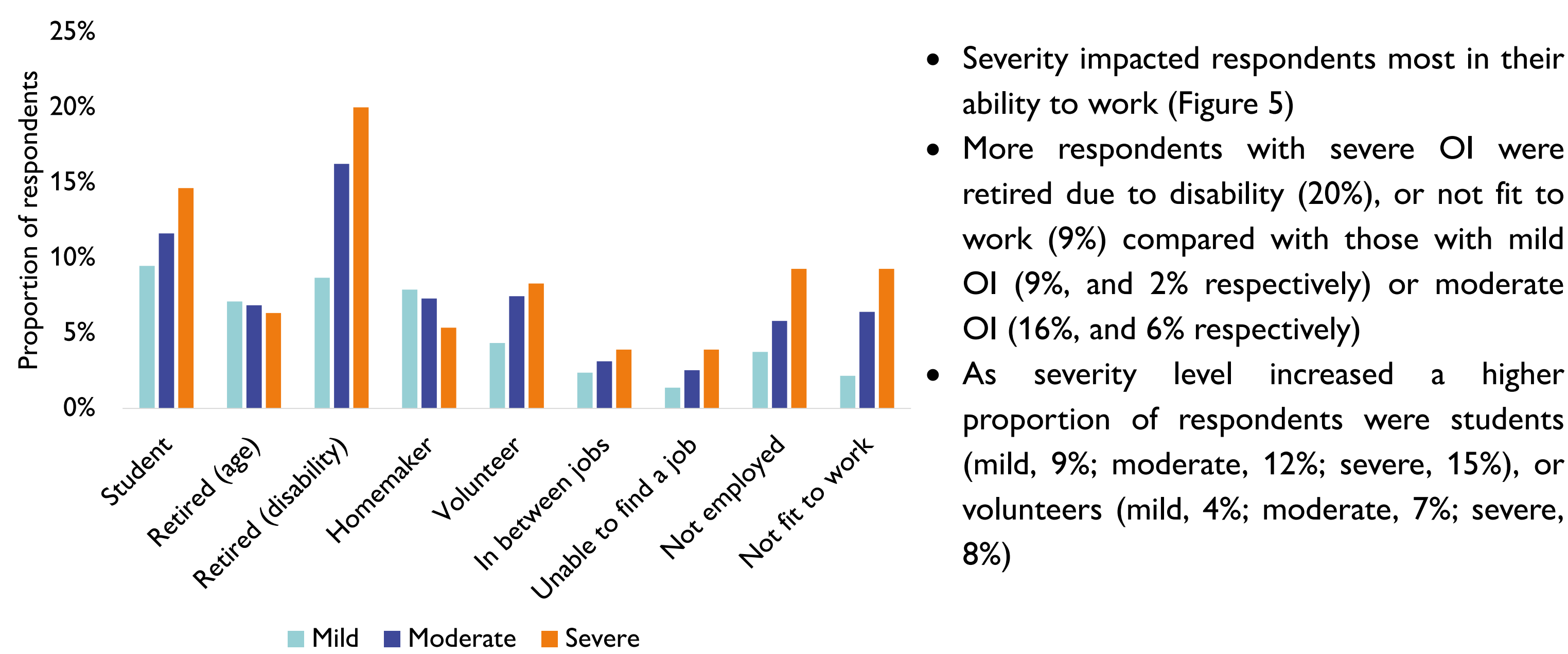
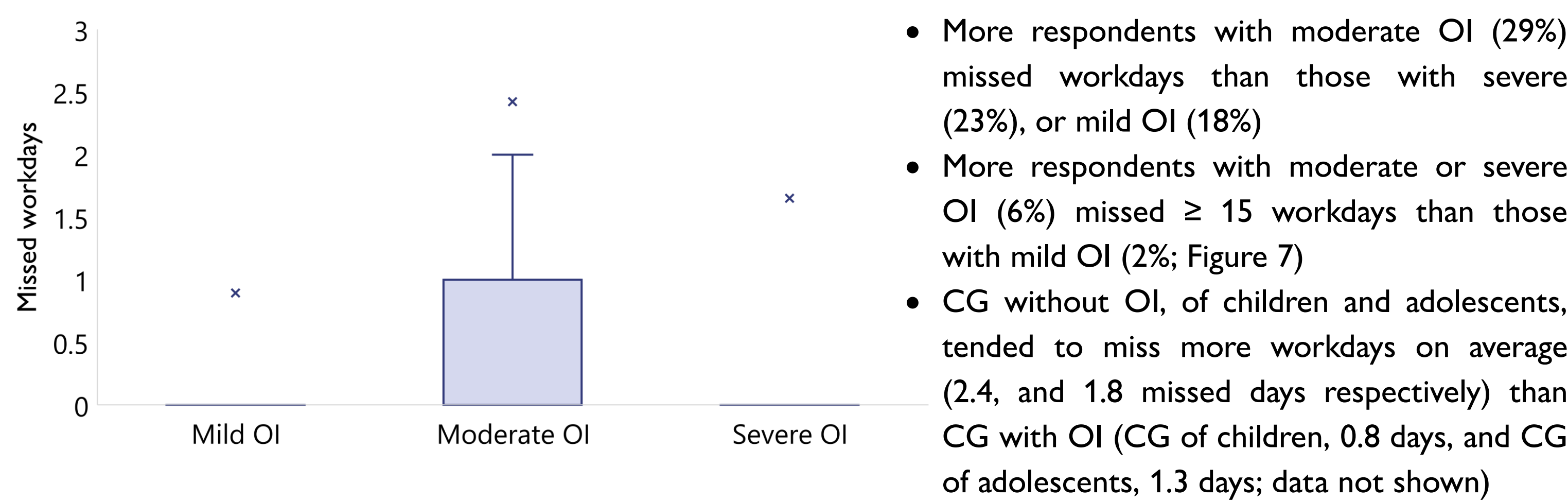


Figure 7. Missed workdays in a 4-week period by OI severity (adults with OI)



Conclusions

- OI affects the productivity of individuals with OI and their CG, limiting their ability to pursue paid employment
- Regardless of CG status or severity levels the number respondents missing workdays is notable
- Increasing OI severity had an increasing impact on paid employment and occupational status, potentially due to increased OI-associated disability
- Improved accommodations may enable some individuals with OI to pursue paid employment, however improved care is needed to address the underlying causes of productivity loss

Acknowledgments

Funding for the IMPACT Survey was provided by Mereo BioPharma Group, London, United Kingdom. The authors would like to thank the OIFE, OIF and national member organisations for their support in the development of the survey and their sustained role in recruiting respondents.

References

1. HOYER-KUHN, H., REHBERG, M., NETZER, C., SCHOENAU, E. & SEMLER, O. 2019. Individualized treatment with denosumab in children with osteogenesis imperfecta - follow up of a trial cohort. *Orphanet journal of rare diseases*, *14*, 219-219