

A global perspective on self-reported productivity losses associated 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 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

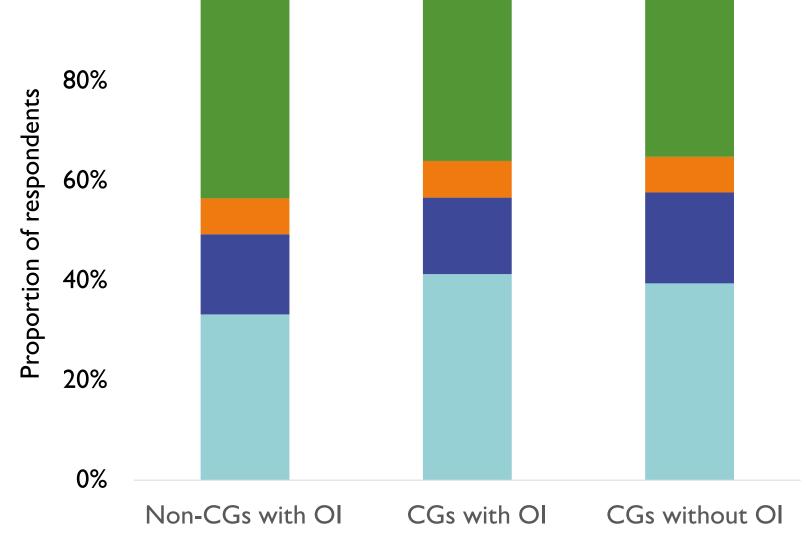
Results

100%

Demographics and respondent characteristics

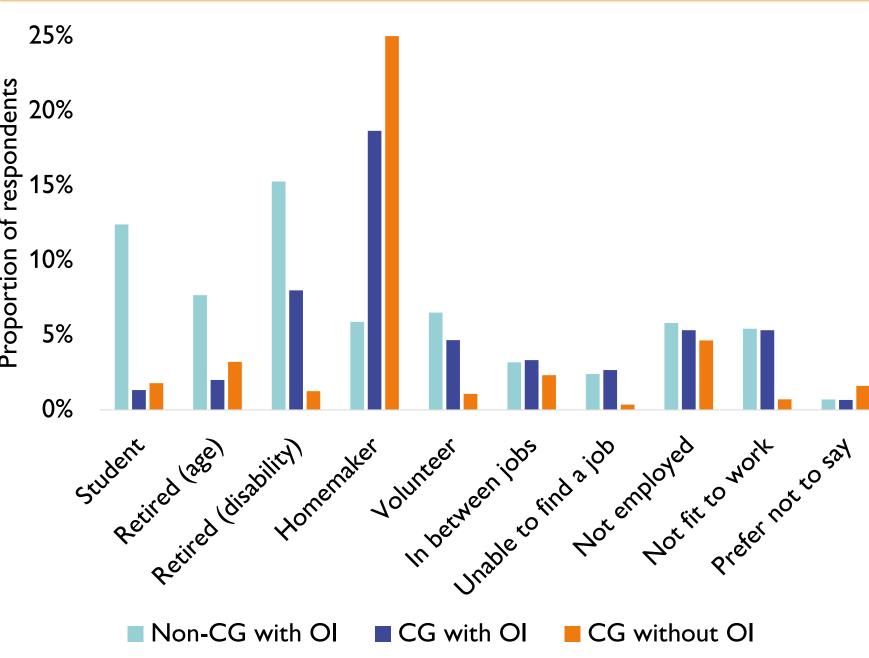
- Overall, 2,208 participants were recruited across 66 countries
- Of 2,000 adult respondents, 1,290 were non-CG with OI, 150 were CG with OI, and 560 were CG without OI (Figure 1)
- Women were more highly represented than men (70% of adults with OI were female, 83% CG without OI were female)
- Most 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



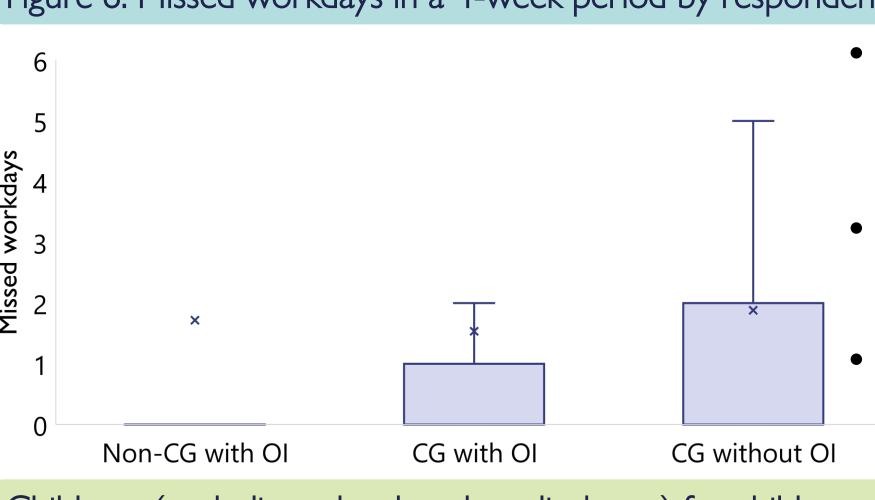
- A lower proportion of non-CG with OI (33%) were employed full time than CG with OI (41%) or CG without OI (39%; Figure 2)
- CG with and without OI had similar levels of full-time employment (41% vs 39%), part-time employment (15% vs 18%), and self-employment (both 7%)
- Employed full timeSelf-employedOther
- Employed part timeNot in paid employment

Figure 4. Occupation other than paid employment by respondent category



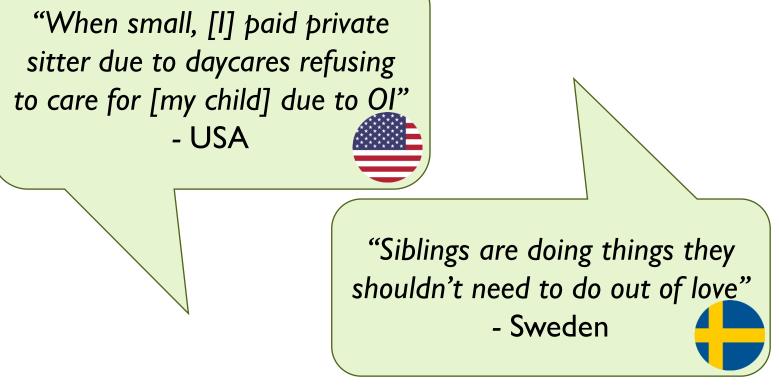
- Occupations undertaken were more similar between CG with and without OI compared to non-CG with OI (Figure 4)
- Retirement due to disability was more common for non-CG with OI (15%) than CG with OI (8%)
- Few respondents were unable to find employment (<3%), or were in between jobs (≤3%)
- 12% of non-CG, and under 2% of CG, with and without OI, were students
- Most CG with (19%) or without OI (25%), were homemakers

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



- Numbers of missed workdays varied for all categories (non-CG with OI, standard deviation [SD] 4.6; CG with OI SD 5.4; CG without OI SD 3.9; Figure 6)
- Non-CG with OI had the highest proportion of respondents (5%) missing ≥ 15 workdays, (2% for CG)
- More CG without OI missed workdays (49%) than CG with OI (30%), or non-CG (23%)

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



References

- 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

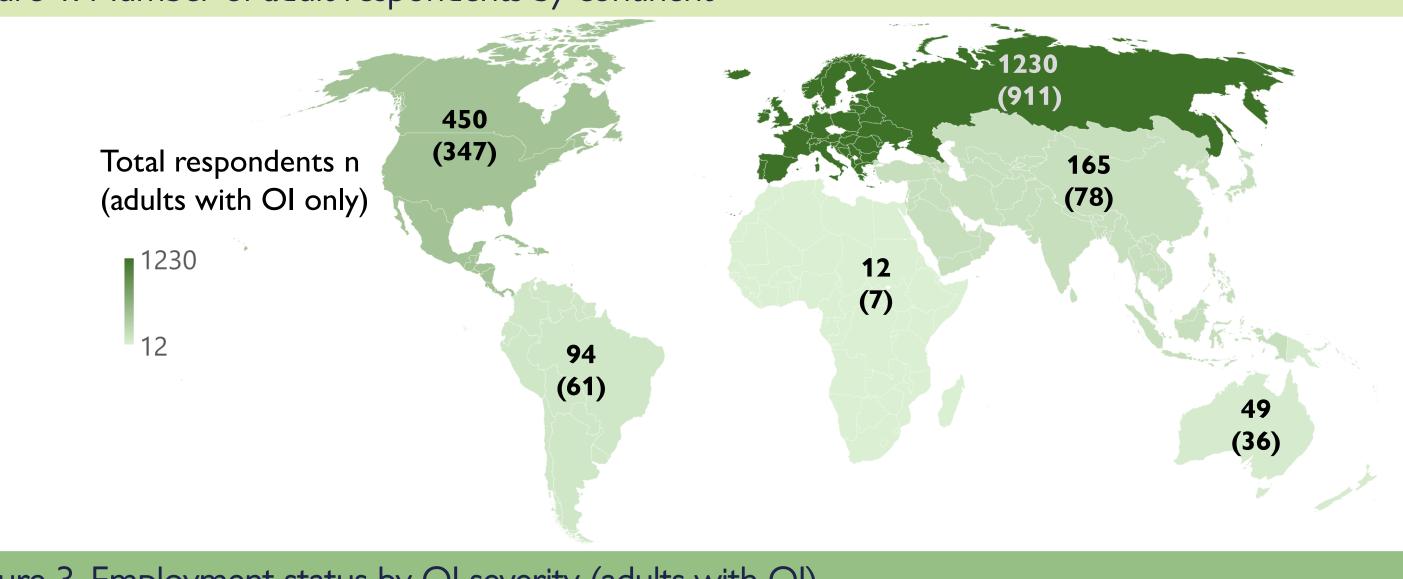
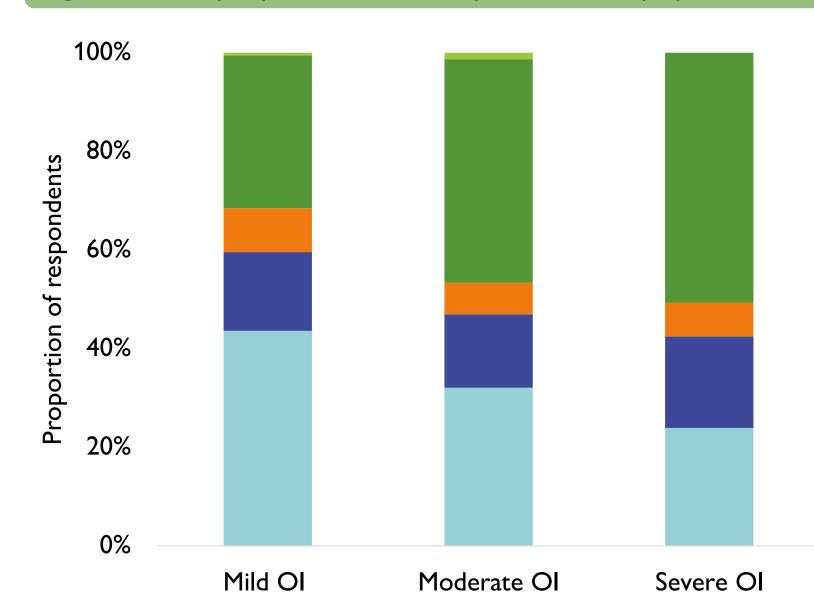
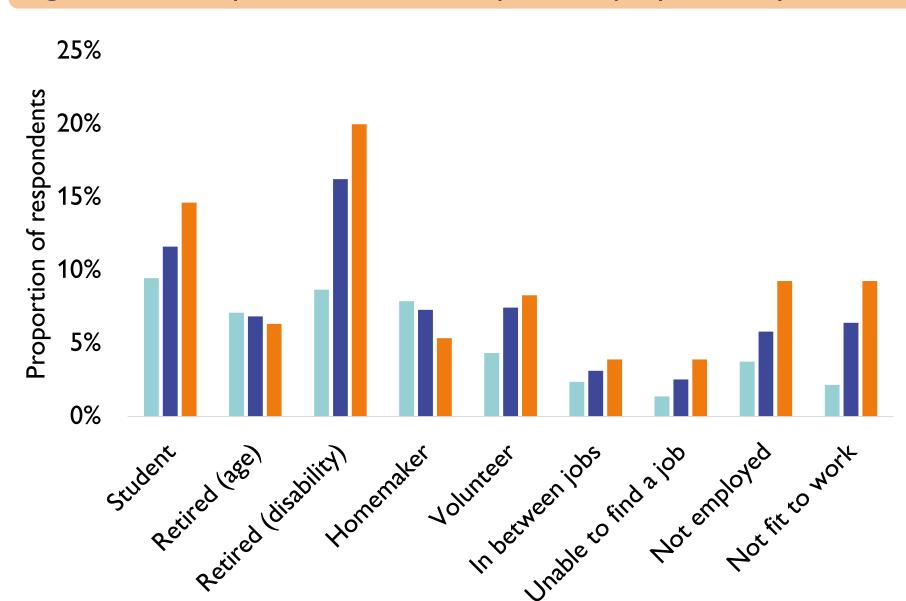


Figure 3. Employment status by OI severity (adults with OI)



- Proportions of people in paid employment decreased with increasing OI severity, due to fewer individuals employed full time (mild OI, 44%; moderate, 32%; severe, 25%; Figure 3)
- Proportions of respondents employed part-time (mild OI, 16%; moderate, 15%; severe, 19%), and self-employed (mild OI, 9%; moderate, 6%; severe OI, 7%) remained similar across severity groups
 - Employed full timeSelf-employedOther
- Employed part timeNot in paid employment

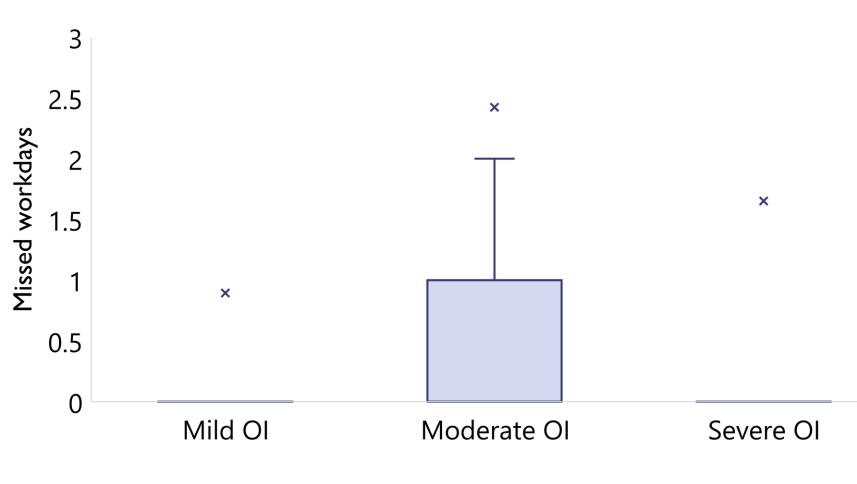
Figure 5. Occupation other than paid employment by OI severity (adults with OI)



- Severity impacted respondents most in their ability to work (Figure 5)
- More respondents with severe OI were retired due to disability (20%), or not fit to work (9%) compared with those with mild OI (9%, and 2% respectively) or moderate OI (16%, and 6% respectively)
- As severity level increased a higher proportion of respondents were students (mild, 9%; moderate, 12%; severe, 15%), or volunteers (mild, 4%; moderate, 7%; severe, 8%)

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

■ Moderate ■ Severe



- More respondents with moderate OI (29%)
 missed workdays than those with severe
 (23%), or mild OI (18%)
- More respondents with moderate or severe
 OI (6%) missed ≥ 15 workdays than those with mild OI (2%; Figure 7)
- CG without OI, of children and adolescents, tended to miss more workdays on average (2.4, and 1.8 missed days respectively) than CG with OI (CG of children, 0.8 days, and CG of adolescents, 1.3 days; data not shown)

Conclusions

- Ol affects the productivity of individuals with Ol 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

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