Disability Identity and Attitudes towards Prenatal Testing in the Osteogenesis Imperfecta Community

Rachel Sullivan1,2, BS; Valentina Pilipenko3, PhD; Nicki Smith3, MS, LGC; Kara Ayers4, PhD
1Department of Human Genetics, Cincinnati Children’s Hospital; 2University of Cincinnati; 3TriHealth; 4University of Cincinnati Center for Excellence in Developmental Disabilities

Background

Osteogenesis Imperfecta (OI) • Genetic skeletal dysplasia that affects 6-7/100,000 births • Leads to short stature and brittle bones that fracture easily

Subtypes • Type I: mild form • Type II: perinatal lethal • Types III, IV, V, and most others: moderate form

Unusually autosomal dominant • Recurrence risk when one parent has OI is 50% • Recurrence risk when neither parent has OI is usually low

Most children with OI are born to parents without OI. Prenatal diagnostic testing is available when OI is suspected based on family history or ultrasound findings. Not all OI is suspected before birth.

Parental Diagnostic Testing (PDT) and the Disability Community

Some disability groups have spoken out about what they feel is a conflict of interests between those providing PDT services and the disability community. Specific disability communities including the achondroplasia and cystic fibrosis communities have reported various attitudes towards PDT.

Disability Identity • Defined as an individual’s view of themselves, their disability, and their connection with the disability community and the larger world.

Limited research exists on disability identity and there are few quantitative evaluation tools.

Objectives

The main aims of this study are to:
1. Describe the disability identity of individuals with OI and parents of children with OI.
2. Describe attitudes families with OI have towards PDT.
3. Evaluate relationship between disability identity and attitudes toward PDT.

Materials and Methods

Questionnaire Outline • Quantitative, cross-sectional, online questionnaire • Demographic information • QDIO: disability identity assessment • Statements assessing attitude toward PDT • Optional comments section

Questionnaire on Disability Identity and Opportunity (QDIO)

• 22 Question assessment of disability identity
• Assesses four factors of identity:
  • Pride
  • Exclusion
  • Social Model
  • Medical Model

• Two versions of QDIO exist:
  • One to measure disability identity in an individual with a disability
  • One to measure disability identity in the parent of a child with a disability.

• Respondents were asked how strongly they agreed with each statement on a scale of 5: Strongly Agree, 4: Agree, 3: Not Sure, 2: Disagree, 1: Strongly Disagree.

PDT Attitudes • Attitudes towards PDT were assessed with two statements:
• If one of my partner were currently pregnant, I would choose to have prenatal diagnostic testing for OI.
  (Personal use attitude)
• Prenatal diagnostic testing for OI should be offered to every pregnant woman routinely.
  (Medical model)

Results

QDIO Results

<table>
<thead>
<tr>
<th></th>
<th>With OI</th>
<th>Without OI</th>
<th>p-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pride</td>
<td>3.280 (0.815)</td>
<td>3.029 (0.815)</td>
<td>0.0437</td>
</tr>
<tr>
<td>Exclusion</td>
<td>2.424 (0.956)</td>
<td>2.503 (0.984)</td>
<td>0.4412</td>
</tr>
<tr>
<td>Social Model</td>
<td>3.779 (0.607)</td>
<td>3.875 (0.604)</td>
<td>0.3240</td>
</tr>
<tr>
<td>Medical Model</td>
<td>3.222 (0.602)</td>
<td>3.360 (0.531)</td>
<td>0.8103</td>
</tr>
</tbody>
</table>

* Significantly different at p<0.05, comparing respondents with OI to those without.

PDT Attitudes

With OI | Without OI
---|---

* Significantly different at p<0.05, comparing respondents with OI to those without.

With OI | Without OI
---|---

* Significantly different at p<0.05, comparing respondents who agree to those who disagree with PDT.

Conclusions and Future Directions

• Families in the OI community are diverse.
• Parents endorsed the medical model over the medical model and pride over exclusion when asked about disability identity.
• Parents with OI were less likely to want to use PDT for their own families than parents without OI.
• Families have many and varied reasons for agreeing or disagreeing with PDT.

Further Direction

• We need more data to define disability identity, including the role of disability identity among nondisabled family and caregivers.
• We need a greater understanding of why individuals within a disability community support or oppose PDT.
• We are hopeful that our research will reveal answers for the OI community and spur further research.

Acknowledgement

• This project is funded in part by a grant from AIDD, ACL, US DHHS P03DD006013-01-OI
• We thank the OI Foundation for their support in recruiting and the OI community for their participation.