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

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Background

Osteogenesis Imperfecta (OI)

- Genetic skeletal dysplasia that affects 6-7/100,000 births
- Leads to short status and brittle bones that fracture easily
- Subtypes
  - Type I: mildest 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 person'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 tool.

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

Participants

- Biological parents who fulfill one or both of the following:
  - Have OI themselves
  - Have a child with OI
- Must be at 18y+ and speak English

Recruiting

- OI Foundation call for research
- OI Foundation social media outlets
- OI Conference Nov. 2017
- Word of mouth

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 I or 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. (Population use attitude)
- Respondents were asked how strongly they agreed with each statement on a scale of 5-Strongly Agree, 4-Agree, 3-Neutral, 2-Disagree, 1-Strongly Disagree.
- We dichotomized data into Agree (4 or 5) and Disagree (1, 2, or 3).

Results

Participant Demographics

	With OI (n=74)	Without OI (n=85)	*p*-value
Age, mean (SD) in years	43 (11.9)	44 (11.4)	0.7508
Race, White, % (n)	96 (71)	92 (78)	0.3402
Sex, female, % (n)	95 (70)	94 (80)	1.0000
Education, bachelor's degree or higher, % (n)	62 (46)	71 (60)	0.2609
Employment, full time, % (n)	39 (29)	49 (42)	0.1959
Respondent, type 1, % (n)	72 (53)	N/A	N/A
***Partner has OI, % (n)**	15 (11)	31 (26)	0.0193
***Has child(ren) with OI, % (n)**	70 (52)	100 (85)	<.0001
***Has >1 child with OI, % (n)**	20 (15)	5 (4)	0.0030

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

Results

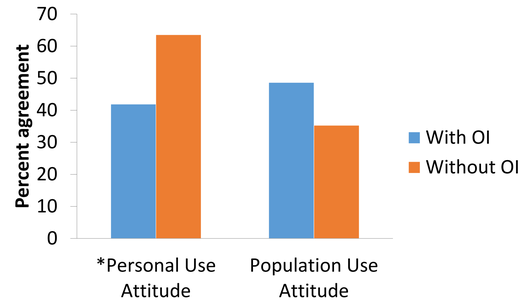
QDIO Results

	With OI Mean (SD)	Without OI Mean (SD)	*p*-value
***Pride**	3.280 (0.815)	3.529 (0.815)	0.0437
Exclusion	2.412 (0.950)	2.303 (0.814)	0.4412
Social Model	3.779 (0.607)	3.875 (0.604)	0.3240
Medical Model	3.282 (0.602)	3.260 (0.531)	0.8103

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

- At a nominal level, respondents without OI reported higher scores in disability pride than respondents with OI. No factors reached statistical significance after multiple testing corrections.
- Pride scores were higher than exclusion scores with $p<0.0001$ for both respondents with OI and respondents without OI. Social model scores were higher than medical model scores with $p<0.0001$ for both respondents with OI and respondents without OI.

PDT Attitudes



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

- For the personal use attitude, 85 (53.5%) respondents agreed. Respondents without OI indicate a stronger agreement (63.5%) than respondents with OI (41.9%) $p=0.0064$.
- For the population use attitude, 66 (41.5%) of respondents agreed. There is no significant difference in agreement between respondents without OI (35.3%) and respondents without OI (48.7%) $p=0.0883$.

Results

Is Disability Identity Associated with PDT Attitudes?

Personal Attitude Use	**Agree**	**Disagree**	**p-value**
Disability Identity Factors	n=85,	n=74,	
Pride, mean (SD)	3.4 (0.8)	3.5 (0.7)	0.3066
Exclusion, mean (SD)	2.3 (0.9)	2.4 (0.9)	0.6115
***Social model, mean (SD)**	3.9 (0.6)	3.7 (0.7)	0.039
Medical model, mean (SD)	3.3 (0.6)	3.2 (0.5)	0.2686
Population Attitude Use	**Agree**	**Disagree**	**p-value**
Disability Identity Factors	n=66	n=93	
***Pride, mean (SD)**	3.6 (0.8)	3.3 (0.7)	0.0455
Exclusion, mean (SD)	2.4 (0.9)	2.3 (0.9)	0.5403
Social model, mean (SD)	3.6 (0.6)	3.4 (0.6)	0.077
Medical model, mean (SD)	3.3 (0.6)	3.3 (0.5)	0.7792

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

- Respondents who endorse of the social model are more likely to agree with the personal use attitude.
- Respondents who endorse pride are more likely to agree with the population use attitude.
- Further analysis is needed to determine causality.

Comments

Fifty-seven respondents left comments on a variety of subjects including explanations of their PDT attitudes:

- "If prenatal testing was routine, we would have known and probably would have avoided a lot of drama on the road to diagnosis."
- "It would help with child abuse cases."
- "Routine prenatal testing would result in many terminations of OI children."
- "… women should be offered testing but they should be well informed on the risks and benefits of testing."

Conclusions and Future Directions

- Families in the OI community are diverse.
- Parents endorsed the social 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.

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