The OI Research Collaborative: Where Researcher and Community Members Connect for a Better OI Life

This presentation was funded through a Patient-Centered Outcomes Research Institute® (PCORI®) Eugene Washington PCORI Engagement Award (14517-OIF).
Overview

1. What is Patient-Centered Outcomes Research (PCOR)?
2. Overview of the OI Foundation’s Grant
3. What the OIF is Currently Doing
4. How you can get involved
Overview of PCOR

• In order to provide the best care and information, the OI Community needs to be driving the science.

• Patient Centered Outcomes Research (PCOR) is different from traditional research. Community members are active participants throughout the entire research process.

• How to get involved

  Determine Questions
  Interpret Results
  How to Conduct a Study
  How to Share Findings
Managing OI: More than Bones

• OI is a disorder of collagen
• Body has 13 different “organ systems”
  *Example*: Skeletal System, Nervous System
• Virtually every organ system in the body contains collagen
  *Example*: Respiratory system

2011 OI Foundation
Online survey of Adult Health Concerns

• OI community expressed on-going problems and concerns about each and every organ system
Impact on quality of life

- Musculoskeletal
  - Current
  - Future
- Hearing
  - Current
  - Future
- Dental
  - Current
  - Future
- Vision
  - Current
  - Future
- Neurologic
  - Current
  - Future
- Gastro-intestinal
  - Current
  - Future
- Pulmonary
  - Current
  - Future
- Cardiologic
  - Current
  - Future
- Urinary
  - Current
  - Future
- Endocrine
  - Current
  - Future
- Skin
  - Current
  - Future
- Oral
  - Current
  - Future
- Hematologic
  - Current
  - Future

mean of scores

0 1 2 3 4 5
Most Concerning: Most health problems were far more common in persons with OI than the general population.
# Skin, Urinary, GI

<table>
<thead>
<tr>
<th>Condition</th>
<th>PREVALENCE</th>
<th>US Population</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>ANHI 2011</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mild (%)</td>
<td>Mod (%)</td>
</tr>
<tr>
<td>Bruises</td>
<td>39</td>
<td>31</td>
</tr>
<tr>
<td>Dry Skin</td>
<td>35</td>
<td>34</td>
</tr>
<tr>
<td>Incontinence</td>
<td>8</td>
<td>10</td>
</tr>
<tr>
<td>Kidney or Bladder Stones</td>
<td>9</td>
<td>13</td>
</tr>
<tr>
<td>Diabetes/High Blood Sugar</td>
<td>11</td>
<td>12</td>
</tr>
<tr>
<td>Low Thyroid Function</td>
<td>8</td>
<td>9</td>
</tr>
<tr>
<td>Constipation</td>
<td>26</td>
<td>27</td>
</tr>
<tr>
<td>Hemorrhoids</td>
<td>17</td>
<td>16</td>
</tr>
<tr>
<td>Reflux/Heartburn</td>
<td>34</td>
<td>34</td>
</tr>
</tbody>
</table>
## Cardiac and Pulmonary

<table>
<thead>
<tr>
<th>Condition</th>
<th>PREVALENCE</th>
<th>US Population</th>
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</thead>
<tbody>
<tr>
<td></td>
<td>ANHI 2011</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mild (%)</td>
<td>Mod (%)</td>
</tr>
<tr>
<td>Blood Vessel Problems (rupture,</td>
<td>5</td>
<td>5</td>
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<td>aneurysm)</td>
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<tr>
<td>Coronary Artery Disease</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>Heart Attack</td>
<td>9</td>
<td>6</td>
</tr>
<tr>
<td>Heart Valve</td>
<td>8</td>
<td>6</td>
</tr>
<tr>
<td>High Blood Pressure</td>
<td>38</td>
<td>33</td>
</tr>
<tr>
<td>High Cholesterol</td>
<td>27</td>
<td>26</td>
</tr>
<tr>
<td>Strokes</td>
<td>5</td>
<td>4</td>
</tr>
<tr>
<td>Cough</td>
<td>16</td>
<td>18</td>
</tr>
<tr>
<td>Shortness of Breath</td>
<td>27</td>
<td>26</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep Apnea</td>
<td>14</td>
<td>14</td>
</tr>
<tr>
<td>Wheezing (Asthma)</td>
<td>16</td>
<td>13</td>
</tr>
</tbody>
</table>
Participants were especially concerned about their future health status in regards to their heart, lungs, vision, endocrine function, and digestive system.
Numerous surveys, focus groups, etc. led to the conclusion that, given the complexity of OI, true understanding of the disorder, and how it changes over time, requires engaging the ENTIRE community.

- And, like the fairy tale, the Little Red Hen, we needed to do it ourselves.
Which led us to...

Apply for and WIN a Patient-Centered Outcomes Research Institute

Eugene Washington Engagement Award
Our Grant

Improving Patient-Centered Outcomes: Expanding Engagement of the Osteogenesis Imperfecta Community

2 years

Launched on June 1, 2019
What is Patient-Centered Outcomes Research?

Research driven by the OI Community’s needs and concerns

OI Community included in entire research process

Results help your doctors address the concerns that you feel are most important to you
Patient-Centered Outcomes Research Engages the Entire Community

People with OI

Parents

Family Members and Caregivers

Doctors and Researchers with an interest in OI
Project Overview

Provide patients, caregivers, and clinicians better information to make important medical decisions.

Develop a coalition of community members and medical professionals who will direct new and inclusive OI research.

Engage the entire OI community in defining important aspects living with OI across the lifespan.
Making Research a Two-Way Street

Old Research Priorities

New Research Priorities
Our Project

Short term:

Collaborate with the entire OI Community, including researchers and doctors, to increase participation of the Community in research

Long Term

Maintain a lasting collaboration with the entire OI Community that leads to better and up to date information with which to make important medical decisions
OK...

So what are we really going to do?
What Are We Doing Right Now?

- Take every opportunity to inform and engage the entire OI community
- You’re the experts! We want to learn from you to better understand the challenges of living with OI to improve research, care, and support

- Regional Meetings
- OIF Newsletters (Research Update coming soon!)
- Fundraising Appeals
- Dedicated OI PCOR website
- National Meeting
- OIF Scientific Meeting
Listen... Recruit .... Practice

- Seek feedback from all events

- Recruit community members to sign up for the OI Patient Registry (a way to communicate with you!)

- Recruit and educate a team of patients/caregivers/clinicians/researchers willing to help guide this work

- Use the OI Patient Registry to begin to explore questions of interest to the community
Tip of the Iceberg

This project will set us up to apply for more grant money to fund projects the OI community cares about
Long Term “Pie in the Sky” Goal

A lasting collaboration with the entire OI Community that leads to better and up-to-date information with which to make important medical decisions across the lifecycle.
Flow of Collaboration

Researchers
- Develop surveys to address issues critical to the OI community

Doctors
- Deliver better personalized care and support for the OI Community

OI Community
- Identify issues critical to the OI community

What does Patient-Centered Outcomes Research mean to a person with OI?

Researchers are better able to understand aspects of living with OI that they hadn’t before.

More research can improve the field of OI care and support.
What does Patient-Centered Outcomes Research mean to a parent of a child with OI?

Parent expertise is considered in research and clinical decision making.

Research can improve the field of OI care and support for your child.
Action Items:

Please Consider Working With Us...

- Look for our articles in Breakthrough
- Follow our updates on our dedicated website (coming soon!)
- Sign up for OI Patient Registry
The most important members ... you!
Committee Leadership

Mark Birdwhistell

Michelle Duprey

Cameron Penn
Additional Steering Committee members

Tracy Hart

Bryce Reeve PhD

Laura Tosi
Do you feel that current research is relevant to you?

Do you feel that researchers understand about living with OI?

Do you talk about your worries to your doctors and find that they’re unable to locate any research that addresses your concern?

Do you feel disconnected from the research being done on OI?

Have you heard of Patient-Centered Outcomes Research?
Thank you!

For any questions, please contact Tracy Hart at thart@oif.org or Rachel Stivers at rstivers@oif.org.