Distribution Agreement

____	____________
Petra Harvey	Date

Identifying Informational Needs of Individuals affected by Osteogenesis Imperfecta and Bridging the Gap to Increase Health Literacy and Patient Engagement

Thesis Report

By

Petra Keturah Harvey, MPH Candidate
Emory Rollins School of Public Health
Executive Master of Public Health Program

____
Iris Smith, PhD, Thesis Advisor
Emory Rollins School of Public Health

____
Tracy Hart, CEO, Field Supervisor
Osteogenesis Imperfecta Foundation

____
Kara Ayers, PhD, Committee Member
University of Cincinnati University Center for Excellence in Developmental Disabilities

____
Reid Sutton, MD, Committee Member
Baylor College of Medicine
Brittle Bone Disorders Consortium

____
Laurie Gaydos, PhD, Associate Chair for Academic Affairs
Emory Rollins School of Public Health

Table of Contents

Chapter 1: Introduction

In the modern culture of information overload, navigating the complex healthcare system can require a tremendous amount of skill and persistence. Many times, patients must piece together information relevant to their condition from a variety of sources, including their physician team, medical brochures or textbooks, support groups, social networking sites, and search engines. The overwhelming amount of general consumer health information available online for the general population can make the task of accessing accurate information difficult. Retrieving, processing, and applying information about a rare health disorder like osteogenesis imperfecta (OI), also known as brittle bone disease, can be a confusing task in the digital era. Twenty years ago, a spokesperson for the US Department of Health and Human Services advised, “Trying to get information from the Internet is like drinking from a firehose, and you don't even know what the source of the water is” (McLellan, 1998). While online healthcare consumers may have physical access through various online sources, the use of jargon and highly technical language can also make information inaccessible. Addressing unmet informational and supportive resource needs is necessary to increase health literacy and patient engagement, and ultimately improve patients’ quality of life and overall health outcomes.

The Evolution of Health Literacy

The definition of health literacy has evolved in many directions since it was first introduced in a 1974 paper about elementary level health education requirements (National Library of Medicine, 2000). Since then, the World Health Organization (WHO), the American Medical Association (AMA), the National Academy of Medicine (NAM), and other health-related organizations and agencies have established several widely accepted definitions. The

NAM definition, which was implemented in *Healthy People 2010*, described health literacy as “the degree to which individuals have the capacity to obtain, process, and understand basic health information and services needed to make appropriate health decisions” (Nielsen et al., 2004). The concern of the overwhelming amount and varying quality of health information available online led *Healthy People 2010* to include a communication objective to avoid significant harm through the spread of inaccurate information (Office of Disease Prevention and Health Promotion, 2000). The consequences of poor health literacy reach far beyond the individual level of the social ecological framework. Beyond improving individually-focused issues like medication adherence or specific care decisions, many health researchers characterize limited health literacy as a “hidden epidemic [that affects] health status, health outcomes, health care use, and health costs” (Siegfried, 2010). Inadequate health and digital literacy skills in the general population are associated with the “inability to assume positive self-management, higher medical costs due to more medication and treatment errors, more frequent hospitalizations, longer hospital stays, more visits to their healthcare provider, and a lack of necessary skills to obtain needed services” (Egbert et al., 2009). Due to its widespread impact of limited health literacy, healthcare organizations and agencies have begun investing in programs and tools to support patient education.

Health Literacy in Rare Disease Populations

Individuals and families affected by chronic rare diseases often face numerous unique challenges in their health journeys. Tasks such as locating knowledgeable physicians, pinpointing an accurate diagnosis and maintaining well-informed multidisciplinary care teams prove to be more and more difficult with disorders that affect less people. Qualitative research studies confirm themes of rare disease patients feeling “forced to become knowledgeable about their own disease state” due to the low prevalence of their medical condition and the scarcity of experienced medical professionals (Budych et al., 2012). Pew Research Center found through a national survey in 2011 that professional support mattered more to patients when the issue involved technical issues such as an accurate medical diagnosis and information about prescription drugs; however, peer-to-peer communication is most helpful in personal issues such as “emotional support in dealing with a health issue or a quick remedy for an everyday health issue” (Fox, 2011). Many patients and advocates turn to community organizations for educational programs, support and resources that complement the information they receive from healthcare providers.

Individuals affected by OI are typically encouraged to connect with the Osteogenesis Imperfecta Foundation (OIF), whose mission is to improve the quality of life for people affected by OI through research to find treatments and a cure, education, awareness, and mutual support. Established in 1970, the OIF has served as a hub for information for the estimated 50,000 children and adults nationwide that live with OI, and the larger community (Hart et al., 2014). Nearly fifty years of offering educational resources and programs tailored to the OI community have been helpful in navigating individual complex decisions, empowering the community, and preparing patients to engage in clinical research.

The decisions that rare disease patients make about their healthcare and treatment plans often require specific and advanced levels of information about their disease and relevant clinical research. Many rare disease patients are often eager to participate in research “hoping to benefit from innovative therapies" (Litzkendork et al., 2016). As with the general population, the levels of need for information and mutual support can vary greatly depending on factors such as access to the internet, health and/or digital literacy, learning styles, format preferences, disease characteristics and family history.

Informational Needs and Sources in the Osteogenesis Imperfecta Community

Common sources of social support and health information organized by the OI Foundation include family conferences and support groups, factsheets, newsletters, podcasts, webinars, and virtual interactive forums. Throughout nearly five decades of service, the OI Foundation "became the first stop for people looking for information on OI because of a new diagnosis, because of a new symptom, or because of a new challenge they are facing" (Hart et al., 2014). Staying in constant communication with constituents gives the OIF unique and valuable insight into the informational needs and sources that the community relies on.

Driven by its mission, the OIF is not only committed to fulfilling programs focused on educational and mutual support, but it also strongly supports research that accelerates a better understanding of OI and improved treatments. The OIF established the OI Adult Natural History Initiative (OI-ANHI) in 2010 to "give voice to the health concerns of the OI community" (Tosi et al., 2015). Led by Dr. Laura Tosi, a long-time member of the OIF's Medical Advisory Council, this research study collected "self-reported disease severity, reported symptoms and health conditions, estimated the impact of [the] concerns on present and future health-related quality of life (QoL) and completed a Patient-Reported Outcomes Measurement Information System (PROMIS®) survey of health issues" (Tosi et al., 2015). Patient-centered outcome research is necessary to produce engaged patient populations and promote the partnership between medical professionals and patients.

Bridging the Gap between Informational Needs and Meaningful Patient Engagement

With the goal of discovering effective therapies and improving health outcomes, it is the objective of healthcare professionals and patient advocacy groups like the OI Foundation to bridge the gaps in knowledge and prepare patients to make informed decisions about their healthcare and future research. Validated tools are available to assess the readability of educational materials and the health literacy levels of a population; however, there is limited evidence to determine the greatest informational needs and concerns of individuals affected by OI. The aims of the present thesis study are threefold:

(1) To assess and describe informational needs within the osteogenesis imperfecta community.

(2) To recommend data-based strategic selection of topics to implement in future OI-related programs and publications.

(3) To identify implications of health literacy and opportunities to increase patient engagement in the osteogenesis imperfecta community and other rare disease patient communities.

The IRB-approved community survey will provide important feedback for the OI Foundation, the Brittle Bone Disorders Consortium, and health professionals who work with the pediatric and adult OI communities. Ultimately, the results of this study intend to enhance strategies to foster health literacy and improve quality of life outcome measures related to a chronic rare health conditions.

Chapter 2: Literature Review

Background

Activities to promote and improve patient-centered education, medical care and research have historically relied on effective communication strategies and partnerships between healthcare providers and patients. In the digital age, the role of the internet and online connections is rapidly changing the formats of communication about health. Health literacy and patient engagement have become critical components of the algorithm for improved health outcomes. These terms, which are typically identified as buzzwords of the healthcare and pharmaceutical industries, have an extensive personal impact in the lives of patients and healthcare consumers. Studies reveal that only 12% of American adults scored in the highest levels of proficiency for health literacy (CDC, 2016). The Nutbeam model of health literacy distinguishes between the following types of health literacy:

1. Basic or functional health literacy, which refers to the reading and writing level needed to understand factual information.
2. Interactive or communicative health literacy, which refers to the skills necessary to apply factual information to new or changing conditions.
3. Critical health literacy, which gauges the ability to critically analyze information to act in the individual's decision-making process (Heijmans et al., 2015).

In addition to the various types of health literacy, the role of digital literacy is also intertwined in patient's understanding of their medical condition and their health outcomes. In the electronic health (eHealth) era, low eHealth literacy levels represent a significant barrier in addressing information gaps (Chan et al., 2011). Health information-seeking behaviors for individuals affected by rare medical conditions are especially of interest to clinicians, health professionals, and patient advocacy organizations. A publication in the Journal of Pediatric Nursing emphasized the importance of "understanding social contexts, including families, peers, everyday activities, and relationships" in patient education and the development of informational materials (Driessnack, 2009). The variable factors involved in improving health and eHealth literacy accentuate the intensive process of developing effective and comprehensive educational materials to enhance patient engagement.

The Shift Towards Internet-Based Consumer Health Information

The internet has become a widely-used medium for sharing information and exchanging ideas related to community resources and coping mechanisms. Online interactive patient communities provide a level of convenience for peer-to-peer education and emotional support. The advantages for the population using internet sources for health information and health literacy may include variable levels of "interactivity, information tailoring, and anonymity", which by design lends itself to disorganized or conflicting recommendations (Cline et al, 2001). With limited resources to promote credible health information and evidence-based consensus recommendations in rare disease populations, virtual forums understandably represent an extremely common method of peer-to-peer support to complement information received from healthcare providers and other resources.

A productive example of the benefits of the cultural shift towards using online medical support forums is a study that mined online social network data for individuals affected by amyotrophic lateral sclerosis (ALS), also known as Lou Gehrig's disease (Nakamura, 2012).

Familial cases of ALS are considered rare based on its prevalence, but the overall awareness of the disease has grown in recent years due to the popular ice bucket challenge (Koohy et al, 2014). The investigators of this study evaluated patient participation on the website PatientsLikeMe (www.patientslikeme.com), which is an online community forum that encourages its members to "track their progress with clinical scales, share information, and learn more about their condition" (Nakamura, 2012). Clinicians and medical researchers interested in ALS have made use of PatientsLikeMe for "a variety of uses including clinical trial recruitment, development of patient-reported outcomes, and perhaps even novel ways of evaluating treatment" (Nakamura, 2012). This study revealed the contributions of understanding a complex and rare disease through the lens of an online patient community forum, especially in the absence of evidence-based data like in other rare disorders. In a *Faster Cures* publication that celebrated five years of patient centricity initiatives, the authors suggest that "real-world evidence comes in the form of patient registries, mobile phone applications, wearable devices, and social media (Faster Cures, 2017).

Although the internet has made general health information and disease-specific patient populations more accessible, this constant availability and sometimes overload has caused a dilemma in the health professional and advocacy communities in their efforts to increase health literacy. Tasks that require advanced health literacy levels are easily translated incorrectly and can cause severe repercussions. For example, understanding the correct timing of a prescription can have drastic consequences, but could easily be misconstrued through a non-reliable information source. Making the decision to participate in clinical research studies is another example of a scenario that could have long-term negative impacts for an individual.

Additionally, individuals who have less money and education are more likely to have insufficient health literacy skills and resort to unreliable information sources, widening the gap in health disparities. Families with less health insurance coverage may look to the internet in desperation to find alternative or less costly treatment options. Awareness and education efforts of voluntary health agencies and patient advocacy groups have begun to address these discrepancies in health literacy and patient engagement research by surveying the knowledge gaps and information sources common in a population.

Knowledge Gaps and Health Literacy Opportunities in Rare Disease Populations

Efforts to promote health literacy must tackle a completely different obstacle in rare disease communities where there is limited data to support professional consensus. A few decades ago, a diagnosis of a rare genetic disease most likely meant that a phenotypically severe child possibly had a shorter average life expectancy. Geneticists and genetic counselors served as critical healthcare providers for rare diseases, as they may have been their first interaction with professional support about a medical condition. As the average life expectancy of OI and other rare disorders increase with the advancement of medicine and technology, patients and their families are tackling the challenge of finding relevant recommendations of an already-scare topic to answer questions over longer life spans. Thus, information for chronic rare disorders must be provided by several types of specialty physicians and tailored to at least two populations, parents of children and adults, who can have drastically different information needs and health literacy backgrounds.

Advancements in diagnostic tools and therapeutic options changed the landscape for rare disease patient education and care. Often limited therapeutic options for rare diseases represent a significant challenge to finding credible and evidence-based health recommendations. The Orphan Drug Act of 1983 rallied more attention and structure to increase the development and approval of therapeutic options (Herder, 2017). The research and advocacy efforts that accompanied the new legislation were substantial, spurring the approval of over 400 products specifically designed for rare disease populations (FDA, 2017). Although there has been progress in the development of new therapies, still 95% of the 7,000 known rare diseases do not have a drug treatment approved by the US Food and Drug Administration (FDA) (FDA, 2017). The lack of approved therapeutic options has significant implications on the ability to translate healthcare information and the availability of practical and proactive messages for rare disease populations. As the Orphan Drug Act of 1983 incentivized the process of developing drugs to treat rare diseases over the years, a number of biomedical publications and pharmaceutical forums have begun to refer to health literacy as the elephant in the patient engagement room (Childs, 2017). Academic and pharmaceutical researchers have come to see patient education as a huge opportunity to increase informed participation in clinical research studies and overall patient engagement.

Patient advocacy groups and researchers involved with conditions that have limited or complex therapeutic options have different strategies to address unmet healthcare and informational needs. Studies on health literacy and patient engagement in populations that have pharmacologic therapy options tend to focus on the implications of unmet informational needs on medication adherence. In the bladder cancer population, a study on informational needs helped assess the type of education and counseling to enhance treatment decision-making aids (Mohamed et al., 2014). This study concluded that differences in demographic and clinical characteristics must be accounted for in the development educational materials and the assessment of a population's needs. The strategies of measuring health literacy and increasing patient engagement must be tailored for each disease-specific community.

Measures of Health Literacy and Models of Information Needs Assessments

Several diagnostic tools have been developed and validated to identify patients with limited health literacy. Two of the most widely used measures include the Test of Functional Health Literacy in Adults (TOFLA) and Rapid Estimate of Adult Literacy in Medicine (REALM) (Nutbeam, 2009). Developed in 1994, the TOFLA measures adult's functional literacy levels in relation to numeracy and reading comprehension of real patient education materials (Parker et al., 1995). The REALM is a similar standardized test that uses common words in word recognition questions, and can be adjusted based on the age and language of the primary target population (Parker et al., 1995). Teach-back or "show-me" methods are common approaches that physicians employ on an individual level to confirm understanding of health information (Williams et al., 2002). Recent research studies have begun to criticize the use of health literacy assessments, emphasizing the need for "more practical and efficient approaches to produce more accessible and easier-to-understand materials" instead of determining new ways to test health literacy (Egbert et al., 2009).

In 2016, a group of German researchers designed a qualitative study to evaluate informational needs for individuals affected by several rare diseases (Litzkendorf et al., 2016). The analysis of conducted interviews defined the following categories: clinical picture, therapy, research, psychological counseling, social law and law-governing benefits, resources, practical information for everyday life, self-help, and comprehensibility of information (Litzkendorf, 2016).

Purpose-Based Information Assessment

While there are limited additional data and publications specific to informational needs within the OI population, there have been studies on similar topics conducted in other populations. Researchers studying informational needs of the prostate cancer population designed a purpose-based information assessment (PIA) to address unmet needs of patients (Feldman-Stewart et al., 2007). Researchers recruited nearly 200 patients and family members to help analyze whether information given to them was more helpful to organize or understand their concerns and treatment options, to make medical decisions or plan treatment, to feel emotionally supported, or to be able to discuss their condition (Feldman-Stewart et al., 2007). Figure 2.1 displays the distribution of reasons patients reported about why they access information about prostate cancer. This study concluded that developing information based on the purposes of consumer use affect “how easily people can search for information and how accurately they process it” (Feldman-Stewart et al., 2007).

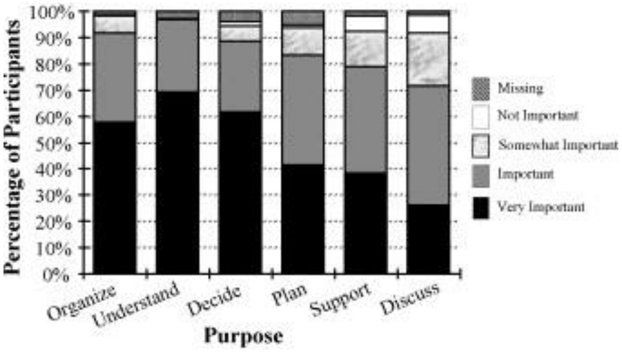


Figure 2.1: Distribution of helpfulness ratings for each purpose (Feldman-Stewart et al., 2007)

Understanding the dynamic needs of the patient, and why they prioritize certain types of health information is an effective assessment method. This approach upholds the concept of patient-centered education, where the value of patient's insights is helpful and necessary to address informational needs.

Strategies for Educational Material Development in Rare Disease Populations

In cystic fibrosis (CF) research, various studies document the importance of emphasizing sensitive and comprehensive information to parents during the newly diagnosed period (Jessup et al., 2016). Jessup's study, which provided an analysis of parental educational experiences following their infant's newborn screening diagnosis of CF, revealed "experiences of shock, disbelief, grief, and a struggle to retain information during this period" (Jessup et al., 2016). Preferences surrounding the ideal amount and timing of information vary greatly depending on the severity of expression; however, parental understanding of the condition and their ability to engage with the care team is vital to influencing their child's outcome. Many study participants also credited experiences meeting "the myriad of people who suddenly entered their world, who were central to the education and support process, and on whom they became immediately reliant" (Jessup et al., 2016). Results of this study indicate and confirm the need for specific subpopulation educational material development.

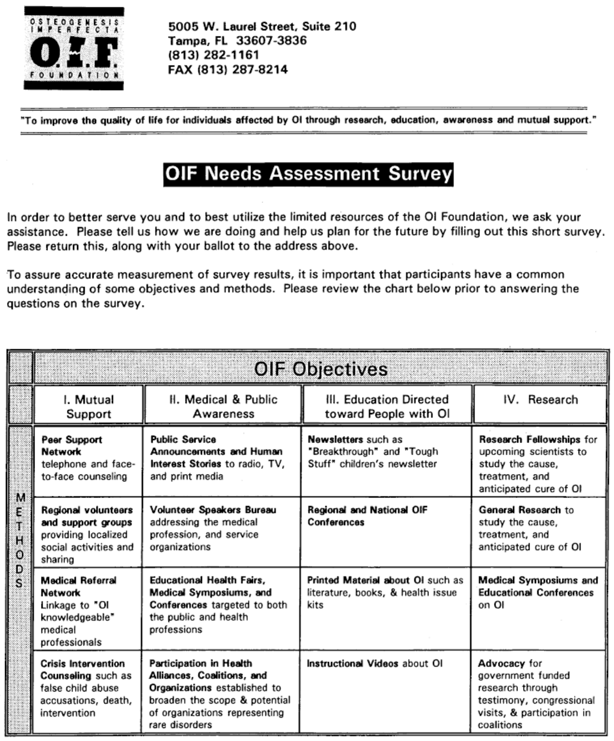
Other studies have suggested stratifying information for the variable informational needs and levels as an effective strategy of comprehensive content development. In a population of the rare disorder congenital hypogonadotropic hypogonadism (CHH), researchers developed a study to assess the readability of CHH-specific educational materials that were co-created by the patient community, clinicians and medical researchers with expertise in CHH. The reading level and readability of these materials were accessed using several validated measures, including:

- “Flesch Reading Ease Formula (evaluates sentence length and number of syllables per word)
- Flesch Kincaid Grade Level (converts the Flesch reading ease formula to a grade level)
- Gunning Fox Index (calculates a weighted average of the number of words per sentence and long words to determine grade level)
- Coleman Liau Index (uses number of characters rather than syllables to determine grade level)
- Simple Measure of Gobbledygook (SMOG, a modification of the Gunning-Fog Index it calculates grade level based on the number of words with 3 or more syllables)
- Automated Readability Index (ratio of difficult words and sentences to provide an estimated age range and grade level)” (Badiu et al., 2017).

The second part of the CHH study asked community research participants to complete the Patient Education Materials Assessment Tool (PEMAT) on educational materials that were co-created by the patient community and medical researchers (Badiu et al., 2017). The PEMAT is an instrument developed by the US Department of Health & Human Services Agency for Health & Research Quality. The authors of this study found that materials co-created by patients “contributed to the high acceptability ratings by patients” (Badiu et al., 2017). They also noted that web-based platforms to distribute informational materials represented effective formats to connect with rare disease patients. This study sets an excellent standard for needs assessments, and has received international accolades for its rigorous approach to the multistep process of assessing a populations needs and developing and evaluating co-created educational materials.

Approaches to Evaluate Informational Needs of the Osteogenesis Imperfecta Community

The complexity of health literacy levels and informational needs in rare disease populations tends to reflect the complexity of the genetic disorder. In an extremely complex and heterogeneous disorder like osteogenesis imperfecta, the lack of informed and experienced physicians can lead to distrust in medical professionals and gaps in knowledge. Limited data exists on informational needs, health literacy and patient engagement specifically in the osteogenesis imperfecta community. Unpublished observations indicate that participation in new research studies is generally overwhelmingly positive in the OI population; however the metrics to gauge this information is largely anecdotal. An OIF Needs Assessment Survey, displayed in Figure 2.2, was designed specifically for the OI



OSTEOGENESIS IMPERFECTA
O.I.F.
FOUNDATION

5005 W. Laurel Street, Suite 210
Tampa, FL 33607-3836
(813) 282-1161
FAX (813) 287-8214

"To improve the quality of life for individuals affected by OI through research, education, awareness and mutual support."

OIF Needs Assessment Survey

In order to better serve you and to best utilize the limited resources of the OI Foundation, we ask your assistance. Please tell us how we are doing and help us plan for the future by filling out this short survey. Please return this, along with your ballot to the address above.

To assure accurate measurement of survey results, it is important that participants have a common understanding of some objectives and methods. Please review the chart below prior to answering the questions on the survey.

	OIF Objectives			
	I. Mutual Support	II. Medical & Public Awareness	III. Education Directed toward People with OI	IV. Research
METHODS	**Peer Support Network** telephone and face-to-face counseling	**Public Service Announcements and Human Interest Stories** to radio, TV, and print media	**Newsletters** such as "Breakthrough" and "Tough Stuff" children's newsletter	**Research Fellowships** for upcoming scientists to study the cause, treatment, and anticipated cure of OI
	Regional volunteers and support groups providing localized social activities and sharing	**Volunteer Speakers Bureau** addressing the medical profession, and service organizations	**Regional and National OIF Conferences**	**General Research** to study the cause, treatment, and anticipated cure of OI
	Medical Referral Network Linkage to "OI knowledgeable" medical professionals	**Educational Health Fairs, Medical Symposiums, and Conferences** targeted to both the public and health professions	**Printed Material about OI** such as literature, books, & health issue kits	**Medical Symposiums and Educational Conferences** on OI
	Crisis Intervention Counseling such as false child abuse accusations, death, intervention	**Participation in Health Alliances, Coalitions, and Organizations** established to broaden the scope & potential of organizations representing rare disorders	**Instructional Videos** about OI	**Advocacy** for government funded research through testimony, congressional visits, & participation in coalitions

Figure 2.2: OIF Needs Assessment Survey
(OI Foundation Archives, n.d.)

community in the early 1990s; however, there is no trackable information on the results of this study (OI Foundation, n.d.). This survey tool asks pertinent questions about methods to advance the OI Foundation's mission in the categories of mutual support, medical and public awareness, education directed toward people with OI, and research (OI Foundation, n.d.). It is unknown by the OI Foundation's current staff and active volunteers whether the survey was distributed, collected, or analyzed. The full four-page survey, which was located in the OI Foundation's Archives, is documented in Appendix A.

The group of physicians and researchers who developed the OI Adult Natural History Initiative (ANHI) sought to address the OI-related health concerns raised by the adult OI population (Tosi et al., 2015). The study administered a set of Patient-Reported Outcomes Measurement Information System (PROMIS®) questions to 959 adult OI patients (Tosi et al., 2015). Developed with funding from the National Institutes of Health in 2004, PROMIS® measures are standardized to assess patient-reported outcomes without clinical interpretation across chronic diseases (Cella et al., 2007). The domains for adults focus on Quality of Life (QoL) related to symptoms of physical, mental, and social health. Tosi's study revealed a "higher prevalence of musculoskeletal and auditory problems and specific QoL concerns regarding the musculoskeletal, auditory, pulmonary, and endocrine systems" in adults living with OI (Tosi et al., 2015). Though this study emphasized the summary of clinical symptoms as reported by adults, there were takeaways that reflected challenges beyond clinical and research settings. One of the most surprising and concerning findings of this study was the statistic that 31% of adults living with OI did not know what type of OI they had (Tosi, 2015). This was a significant revelation in the understanding of basic informational needs and health literacy levels in the OI population.

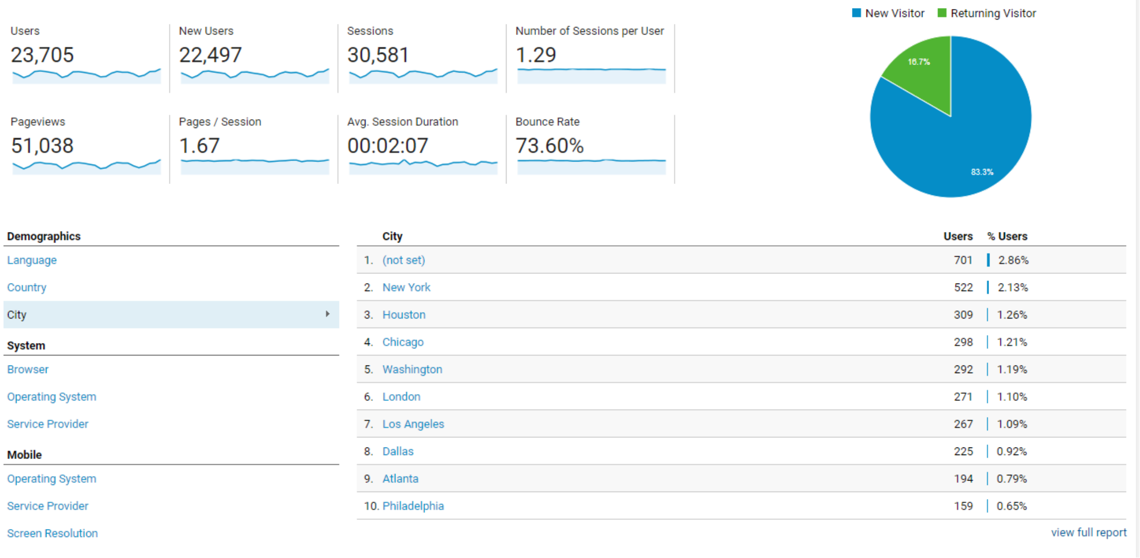
QoL studies have become extremely common in chronic rare diseases, especially those with potential therapies moving toward clinical trials. Studies that rank the top health-related QoL concerns from the firsthand experiences of adults living with OI undoubtedly play a necessary role in clinical practice, research, and drug development; simultaneously, it represents a tremendous service to the community. They also provide valuable insight into the patient experience which the OI Foundation can apply to developing effective programs, publications, advocacy priorities and decision-making tools. As a direct outcome of this study, the OI Foundation was able to implement multiple outreach efforts under the "Adult Health Initiative" and publish the digital and print "Take Charge of Your Health Toolkit" (OI Foundation, 2017). The products of this study have been enthusiastically received by the OI community. The OI Foundation continues to advocate strongly for research that promotes a better understanding of OI and translates to meaningful health information for constituents.

Alternative Means of Characterizing the Osteogenesis Imperfecta Community

Prevalence estimates of osteogenesis imperfecta in the United States range from 25,000 – 50,000 (Tosi et al., 2015). The wide range of prevalence of OI can be explained by several reasons, including the variable numbers of an extremely mild undiagnosed population, the difference in diagnostic methods across physicians and hospitals, and the "phenotypic expansion" that many rare genetic disorders have seen over the years with next-generation sequencing (Patel et al., 2015). Population and program metrics are critical for the OI Foundation to implement relevant and effective activities. Since the establishment of the Brittle Bone

Disorders Consortium (BBDC), the OI Foundation monitors demographics as reported from the BBDC Contact Registry to tailor subject-specific or subpopulation-specific materials and activities.

Many companies have maximized the use of free search engine optimization (SEO) tools to characterize their constituent population. The OI Foundation uses Google Analytics to monitor the demographics of website visitors. According to the Google Analytics page for the OI Foundation website (www.oif.org), there is an average of approximately 23,000 monthly website visitors (Google Analytics, 2018). Figure 2.3 displays the February 2018 report, which includes a list of the top cities from which individuals access the website. Of the cities listed, London is the only non-US city that ranks in the top ten (Google Analytics, 2018).



Users
23,705

New Users
22,497

Sessions
30,581

Number of Sessions per User
1.29

Pageviews
51,038

Pages / Session
1.67

Avg. Session Duration
00:02:07

Bounce Rate
73.60%

Demographics
Language
Country
City

System
Browser
Operating System
Service Provider

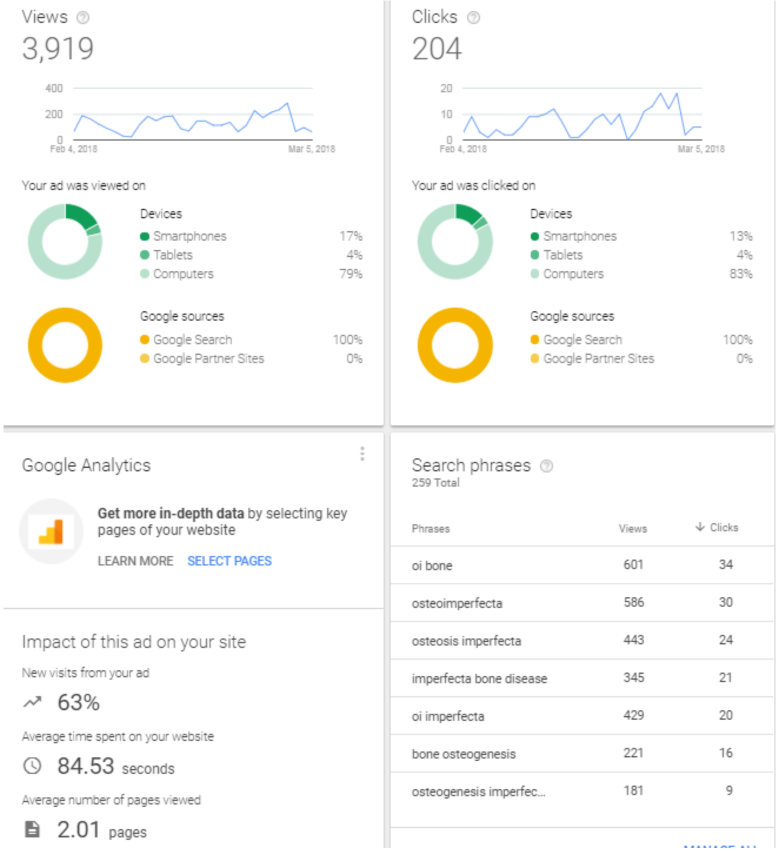
Mobile
Operating System
Service Provider
Screen Resolution

	City	Users	% Users
1.	(not set)	701	2.86%
2.	New York	522	2.13%
3.	Houston	309	1.26%
4.	Chicago	298	1.21%
5.	Washington	292	1.19%
6.	London	271	1.10%
7.	Los Angeles	267	1.09%
8.	Dallas	225	0.92%
9.	Atlanta	194	0.79%
10.	Philadelphia	159	0.65%

view full report

Figure 2.3 Google Analytics Page for OI Foundation Website, Example Report (Google Analytics, 2018)

Figure 2.4 is an example of a report of common related search phrases, the average time spent on the website, the average number of pages viewed, and the types of devices (computer, mobile, or tablet) used to click on a Google Ad for the OIF (Google Analytics, 2018).



Views
3,919

Your ad was viewed on

Devices	
● Smartphones	17%
● Tablets	4%
● Computers	79%

Google sources	
● Google Search	100%
● Google Partner Sites	0%

Clicks
204

Your ad was clicked on

Devices	
● Smartphones	13%
● Tablets	4%
● Computers	83%

Google sources	
● Google Search	100%
● Google Partner Sites	0%

Google Analytics

Get more in-depth data by selecting key pages of your website

LEARN MORE SELECT PAGES

Impact of this ad on your site

New visits from your ad
63%

Average time spent on your website
84.53 seconds

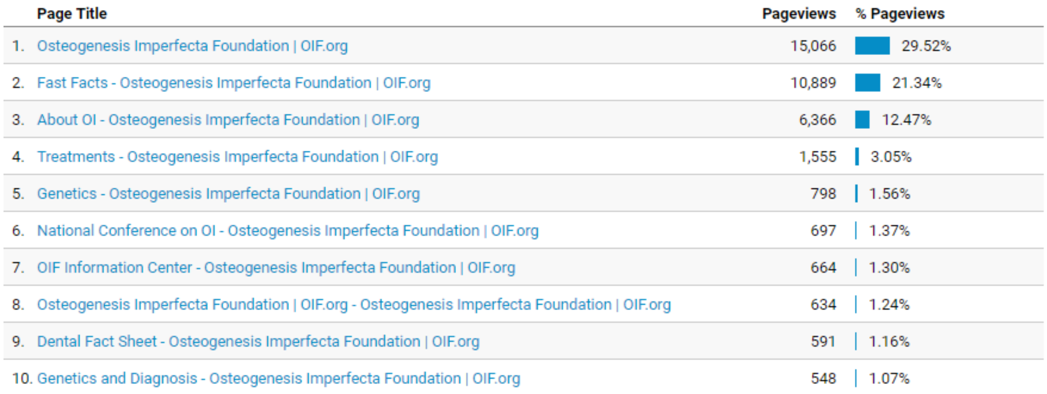
Average number of pages viewed
2.01 pages

Search phrases
259 Total

Phrases	Views	↓ Clicks
oi bone	601	34
osteoimperfecta	586	30
osteosis imperfecta	443	24
imperfecta bone disease	345	21
oi imperfecta	429	20
bone osteogenesis	221	16
osteogenesis imperfec...	181	9

Figure 2.4 Google Analytics Page for OI Foundation Google Ad.
(Google Analytics, 2018)

Another helpful report from Google Analytics identifies the most frequently visited webpages. Figure 2.5 lists the top ten most viewed webpages within the framework of the OIF website.



	Page Title	Pageviews	% Pageviews
1.	Osteogenesis Imperfecta Foundation | OIF.org	15,066	29.52%
2.	Fast Facts - Osteogenesis Imperfecta Foundation | OIF.org	10,889	21.34%
3.	About OI - Osteogenesis Imperfecta Foundation | OIF.org	6,366	12.47%
4.	Treatments - Osteogenesis Imperfecta Foundation | OIF.org	1,555	3.05%
5.	Genetics - Osteogenesis Imperfecta Foundation | OIF.org	798	1.56%
6.	National Conference on OI - Osteogenesis Imperfecta Foundation | OIF.org	697	1.37%
7.	OIF Information Center - Osteogenesis Imperfecta Foundation | OIF.org	664	1.30%
8.	Osteogenesis Imperfecta Foundation | OIF.org - Osteogenesis Imperfecta Foundation | OIF.org	634	1.24%
9.	Dental Fact Sheet - Osteogenesis Imperfecta Foundation | OIF.org	591	1.16%
10.	Genetics and Diagnosis - Osteogenesis Imperfecta Foundation | OIF.org	548	1.07%

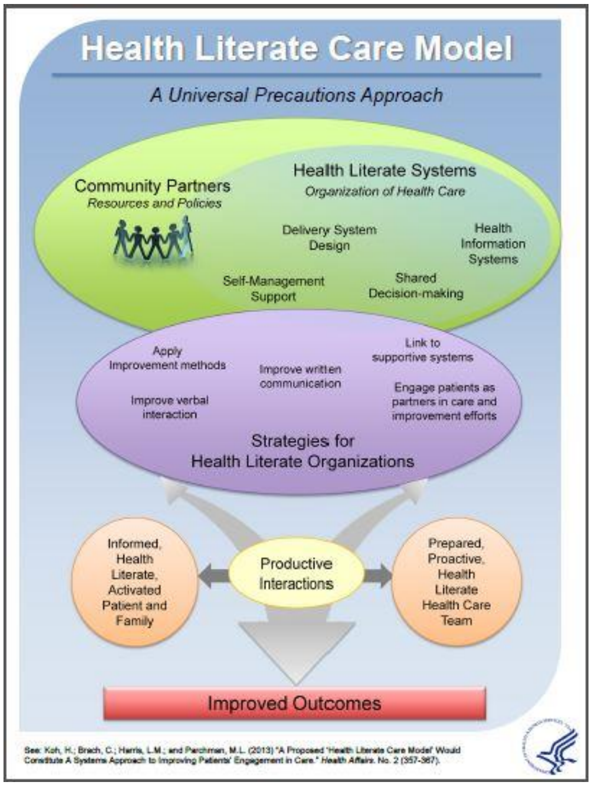
Figure 2.5 Google Analytics Page for OI Foundation’s Most Viewed Webpages (Google Analytics, 2018)

Google Analytics is a powerful tool that can save time for small organizations. Facebook and many other social media sites also provide activity insight reports that summarize page views, page likes, reach, and other statistics. Free access to customizable data significantly strengthens the OIF communication strategy for online information materials.

Increasing Health Literacy through Internet-Based Sources of Information

The OIF website and Facebook page may be the primary sources of information available on the internet, but they are far from the only sources. There are hundreds of interactive online pages and forums that may be relevant to the OI community. Community members may experience advantages and disadvantages with expanded internet access as sources of information about OI. The 2011 Pew Research Center national survey found that internet forums have been helpful in the diagnostic odyssey of populations with complex inheritance or transmission patterns (Fox, 2011). One survey respondent to this survey commented, “When time is of the essence, search on the internet is the only way to time travel” (Fox, 2011). While many people find the internet to be a valuable supplement to their health care needs, the risk of misinformation is concerning. With the extreme pace of social networking sites, the risk of spreading misinformation about complex and sometimes severe disorders is exceptionally dangerous. Rapid advances in scientific knowledge make it easy to find comfort in the pace of online forums in the eHealth era; however, unreliable information can be counterproductive. Information sources such as Wikipedia, Facebook, and other popular yet, uncertified sites can exacerbate confusion, frustration and low health literacy levels.

The Health Literate Care Model (Figure 2.6) was developed by the Agency for Healthcare Research and Quality (AHRQ) (Scotten, 2015). The diagram displays the critical link “between community partners, health literate systems, and health literate organizations as they interact productively with each other to improve outcomes” (Scotten, 2015).



See: Koh, H.; Brach, C.; Harris, L.M.; and Parchman, M.L. (2013) "A Proposed 'Health Literate Care Model' Would Constitute A Systems Approach to Improving Patients' Engagement in Care." Health Affairs. No. 2 (357-367).

Figure 2.6 Health Literate Care Model
(Scotten, 2015)

With the scarcity of evidence-based development of health information for rare diseases and the lack of knowledgeable medical professionals, factors outlined in the research studies and example assessments previously described must be considered by patient advocacy groups and healthcare providers in the process of developing educational materials and support resources. The strategic approach of co-creating patient education materials reaffirms the value of involving OI patients and community members in all steps of the research pathway and process to develop effective educational materials. An understanding of the information and support needs of disease-specific populations is vital to progress in OI clinical care and research in the 21st century.

Insights gained from this thesis project will strengthen the design of educational resources of the OI Foundation, as well as the programs and publications of the Brittle Bone Disorders Consortium, and other relevant US government health agencies and patient advocacy groups. Further research is critical to develop data and understand the gaps in knowledge, informational needs and health literacy levels in populations affected by chronic rare genetic disorders such as osteogenesis imperfecta.

Chapter 3: Methods and Student Contribution

Primary data collection was required for this thesis project. The evidence from informational needs assessments in other disease-specific populations served as rationale to develop a 27 question OIF Program Services Survey for the osteogenesis imperfecta community. Feedback from the OIF's Board of Directors and Medical Advisory Council were incorporated prior to submission to Emory University's Institutional Review Board (IRB). Petra Harvey was the Principal Investigator. With the guidance of her Thesis Advisor Dr. Iris Smith, Field Advisor OIF Chief Executive Officer Tracy Hart, and Thesis Committee Members Dr. Reid Sutton and Dr. Kara Ayers, Petra completed the following activities:

- Designed survey instrument (Appendix B)
- Secured ethics approval (Appendix C)
- Disseminated recruitment messages through mail/email/social media (Appendices D, E)
- Organized data and analyzed results
- Developed figures and tables
- Prepared thesis report, manuscript for a peer-reviewed journal, and executive summary for the OI Foundation and Brittle Bone Disorders Consortium.

Petra intends on submitting a manuscript to Elsevier's Patient Education and Counseling Journal.

Survey Instrument Design

Based on evidence from published needs assessments in other disease-specific populations, a 27 question Program Services Survey was developed for the osteogenesis imperfecta community. A report on evaluating an educational website for adults living with cystic fibrosis provided examples of disease-specific questions regarding patient education materials and sources (Anand et al., 2013). A different study on information-seeking behaviors in inherited bone marrow failure syndromes was also reviewed in the process of defining categories for socio-demographics and health information sources (Hamilton et al., 2015). A mix of multiple-choice, multiple-selection, and Likert rating scale questions were included. The target audience of the survey instrument included OIF constituents and individuals over 18 years of age who are affected by OI. Feedback from the OIF’s Board of Directors and Medical Advisory Council were incorporated in the content and design. The suggested categories of sociodemographic data included sex, age range, connection to OI, and the type of OI (self or connection). Race, ethnicity, geographic location, education background, and income level were sociographic categories not included to preserve anonymity. The survey template and introduction letter were printed in lay language and mirrored on the Survey Monkey platform for online access.

Ethics Consideration

This study had minimal risks, and was declared exempt by the Emory University Institutional Review Board on January 30, 2018 (Appendix C). Informed consent was prompted for online and paper survey respondents before questions were presented, and participants were assured of anonymity and confidentiality.

Recruitment and Data Collection

Participant recruitment activities included an email announcement (Appendix D), mailed paper surveys (Appendix B), and social media announcements (Appendix E) with instructions on how to complete the survey. Four reminders were disseminated through email and social media (Appendix E). The survey was administered via both the Survey Monkey platform and a mailed paper form. Data was collected over a three week period in February 2018, including a pre-planned one week extension added on to the original two week timeline.

Data Analysis

The target goal for survey participation was 300. Of the 400 online and paper survey responses collected over three weeks, 15 respondents indicated they did not wish to be counted in the analysis, leaving a total of 385 responses (see Figure 3.1).

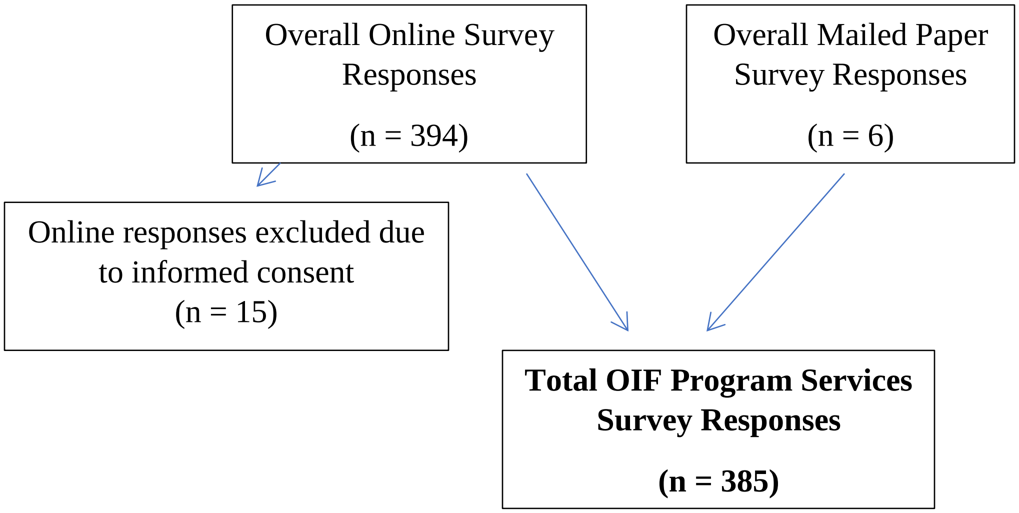


Figure 3.1 OIF Program Services Survey Respondent Flowchart

Frequency and cross-tabulation statistics were generated for raw quantitative data. Write-in comments and responses were analyzed and coded according to themes to avoid redundancy.

Chapter 4: Results

After manually adding in the de-identified and confidential responses from mailed-in paper survey forms and deleting online responses from individuals who did not wish for their responses to be included in the analysis, a total number of 385 participant responses were evaluated. According to Survey Monkey, the OIF Program Services Survey had a 77% completion rate and took online respondents an average of five minutes to complete. Respondents were permitted to skip responses to any question, so the number of responses (n) varies for each question or category.

Survey Respondents Demographics

Most participants (84%) who completed the survey were women. The largest age category self-reported among survey respondents (40%) was ages 36-54 years old, followed by 55-72-year-old respondents, who represented 38%. The majority of respondents (58%) reported that they had OI themselves. Forty-four percent of respondents reported they were parents of individuals with OI. The remaining categories of respondents' connections to OI varied: 14% were family members of an individual with OI, 8% were grandparents of an individual with OI, 7% were health professionals, 4% were spouses married to a person with OI, 1% of respondents were friends of an individual with OI. The categories of respondent's connections to OI were presented in a check-box response question, so respondents could select all that applied.

The full analyses of demographics collected on this survey are outlined in Table 4.1.

Table 4.1
Survey Respondent Demographics (overall n = 385)

Category	**n (%)**
Sex	**309**
Male	45 (14.56)
Female	260 (84.14)
Prefer Not to Disclose	4 (1.30)
Age	**307**
18-36 years old	50 (16.29)
36-54 years old	124 (40.39)
55-72 years old	116 (37.79)
73 or older	13 (4.23)
Prefer Not to Disclose	4 (1.30)
Connection to OI	**308**
Self	179 (58.12)
Parent/Guardian	134 (43.51)
Spouse	11 (3.57)
Grandparent	26 (8.44)
Family Member	44 (14.29)
Friend	4 (1.30)
Health Professional	21 (6.82)
Other	16 (5.20)
Type of OI (Respondent or Connection)	**307**
Mild	125 (40.72)
Moderate	109 (35.50)
Severe	69 (22.48)
Not Sure	23 (7.49)
Not Applicable	7 (2.28)
Prefer Not to Disclose	1 (0.33)

Information Materials and Resources

The five highest rated topics for OI-related informational materials and resources included: bone/orthopedic (65%), pain management (46%), adult general health (45%), hearing loss (30%), and dental care (30%). Table 4.2 shows the subject areas by topic and their corresponding responses.

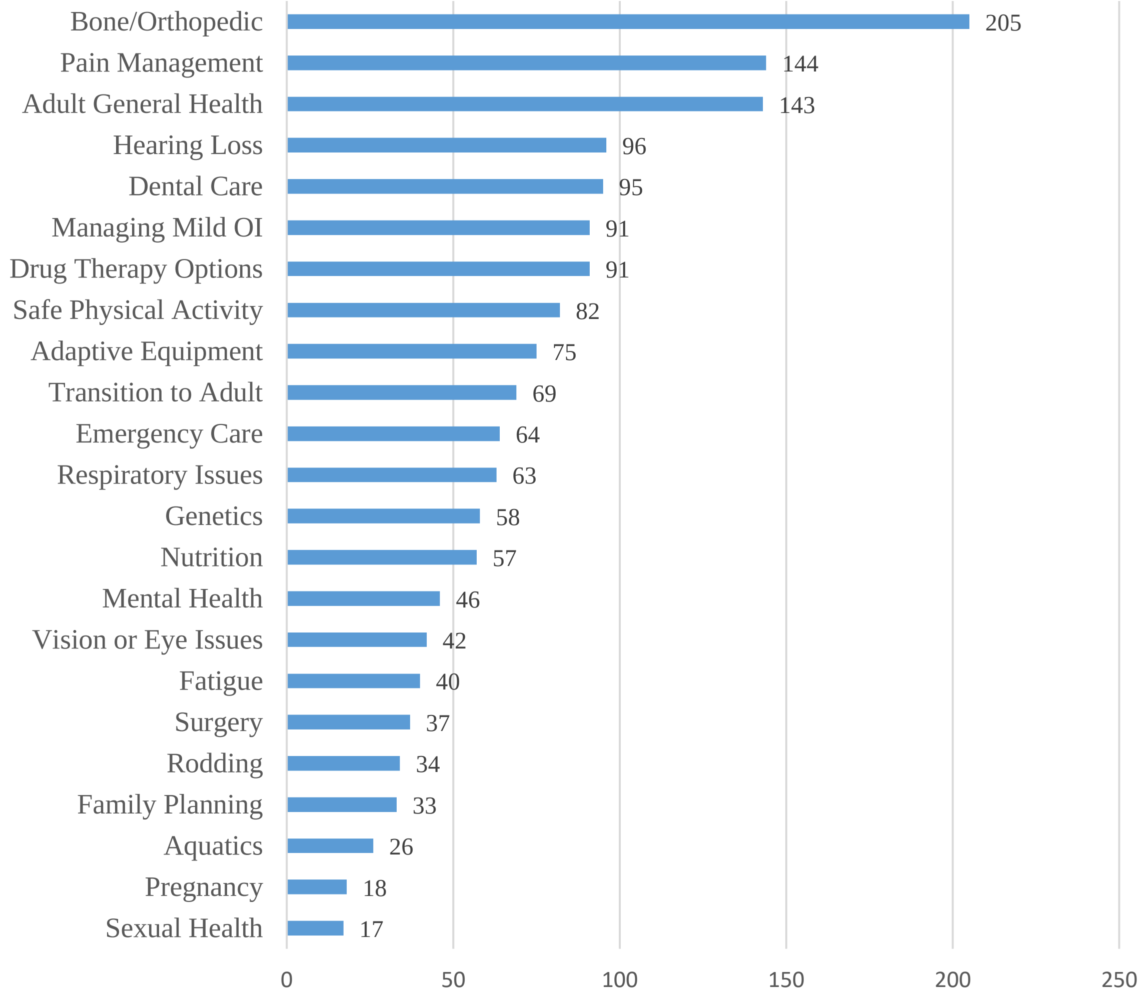
Table 4.2 Numeric Data
Question: Of the subject areas listed below, which are your five most important topics for OI-related informational materials and resources. (n = 316)

Subject Areas	**n (%)**
Bone/Orthopedic	205 (64.87)
Pain Management	145 (45.89)
Adult General Health	143 (45.25)
Hearing Loss	96 (30.38)
Dental Care	95 (30.06)
Managing Mild OI	91 (28.80)
Drug Therapy Options	91 (28.80)
Safe Physical Activity	82 (25.95)
Adaptive Equipment	75 (23.73)
Transition to Adult	69 (21.84)
Emergency Care	64 (20.25)
Respiratory Issues	63 (19.94)
Genetics	58 (18.35)
Nutrition	57 (18.03)
Mental Health	46 (14.56)
Vision or Eye Issues	42 (13.29)
Fatigue	40 (12.66)
Surgery	37 (11.71)
Rodding	34 (10.76)
Family Planning	33 (10.44)
Aquatics	26 (8.23)
Pregnancy	18 (5.70)
Sexual Health	17 (5.40)

Figure 4.1 displays a more visual analysis of the most important topics for informational materials and resources.

Figure 4.1

Question: Of the subject areas listed below, which are you five most important topics for OI-related informational materials and resources? (Please select five.)



Twenty-three write-in responses were analyzed for this question. The additional suggested topics for OI-related informational materials and resources are grouped by themes in Table 4.3.

Table 4.3 Write-In Responses **Question: Of the subject areas listed below, which are your five most important topics for OI-related informational materials and resources.**
Medical Topics
Physical Therapy
Cardiac issues, heart health
Surgery and risks for adults
Joint laxity issues in later life
Managing the associated onset of arthritis
Effects of menopause on OI
Pre-diagnosis and testing options
OI Type II
Age-Related Topics
Adolescence, Transition
Advanced/premature aging and early death
Increase of fractures as one ages
Prognosis past teen years
Non-Medical Topics
Mental- treating the whole person
How to get involved with research
Healthcare options
Socialization for adults with OI
Advice on how to persuade insurance companies of the need for adaptive equipment
Vigorous exercise (i.e. what is considered too much?)
Recent books published with OI as subject or part of

Table 4.4 summarizes information about how often respondents visit the OIF website. Most respondents (53%) visit the website approximately once every couple of months. Other respondents visit the website more frequently, either once or twice a month (33%) or three to five times per month (13%).

Table 4.4
Question: Approximately how often do you visit www.oif.org? (n = 274)

3-5 times per month	36 (13.14)
1-2 times per month	91 (33.21)
Once every 2 months	145 (52.92)

Forty-one respondents selected "other" and wrote in responses to describe how often they visit the OIF website. Table 4.5 is a summary of unique write-in responses.

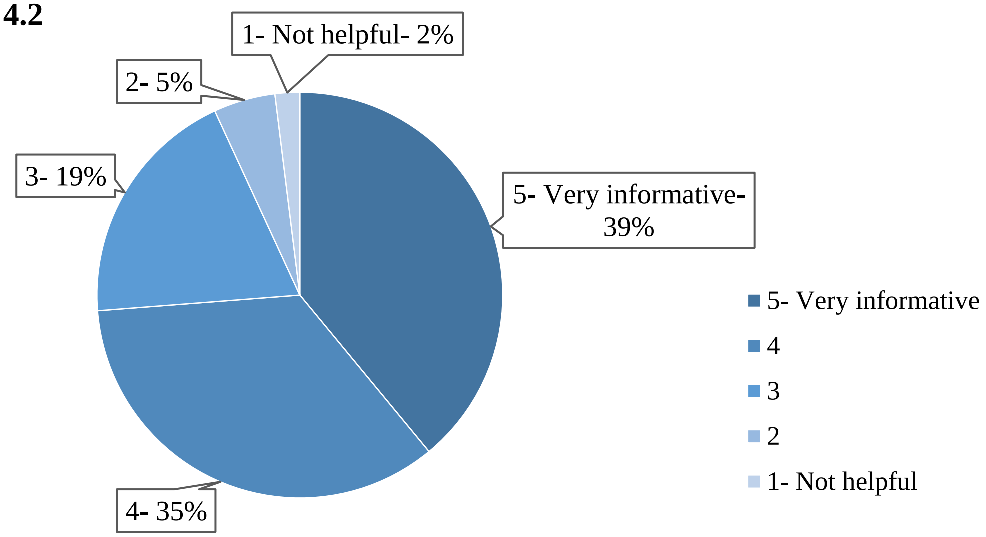
Table 4.5 Write-In Responses
Question: Approximately how often do you visit www.oif.org?

Once a year or less
Twice a year
About once a year
Not often
Rare
Never
About once a quarter
Variable
Sporadically
As needed, more often during convention planning
When I need to register for something
My child is grown so I am less involved
Parent of grown child with OI, not often
Whenever I get an email from oif.org or see a Facebook post
Not very often since I was not diagnosed until my early 50s after menopause.
New to OIF

On a Likert scale of 1 (not helpful) to 5 (very informative), survey participants were asked to rate the quality of OIF materials and resources. 119 of 305 respondents (39%) found OIF materials and resources very informative. The weighted average of 305 responses was 4.04/5. The breakdown of responses is displayed in Figure 4.2.

Question: Please rate the quality of OIF materials and resources.

Figure 4.2



Fifteen respondents wrote-in their responses for this question. Table 4.6 is a summary of positive critiques and constructive suggestions.

Table 4.6 Write-In Responses **Question: Please rate the quality of OIF materials and resources.**
Positive Critiques
Great lay information
I have recently had problems with my teeth so I went to find dental care information and got a lot of great information from the site.
Suggestions
Materials are too medically written
I wish there was more information on the effectiveness of various drug therapies for adults

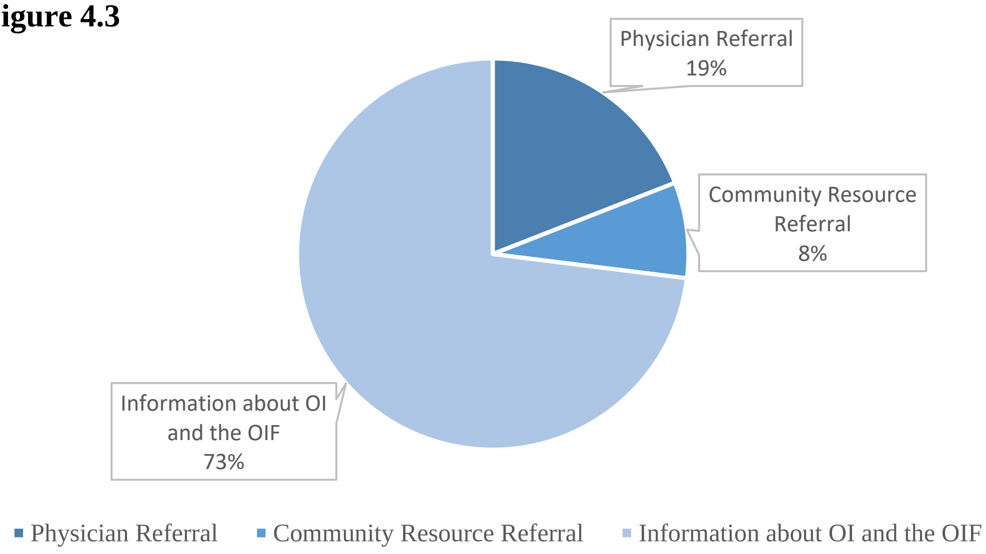
Often Google provides outdated results- some old URLs and PDFs are still available.
Following the website menu to what I want can be challenging
Not much available on aging, would like to see more adult-oriented information
Lacking in pain management information for elderly
They are vague to cover all types of OI, making it less useful

Information Center Services

Figure 4.3 summarizes the proportion of respondents who have utilized OIF Information Center services or resource by calling or emailing the OIF Office. Of the 234 survey participants who responded to this question, 73% reported they had received information about OI and the OIF, 19% reported they have received a physician referral, and 8% reported they have received a community resource referral.

Question: Which services or resources have you utilized by calling or emailing the OIF Office?

Figure 4.3

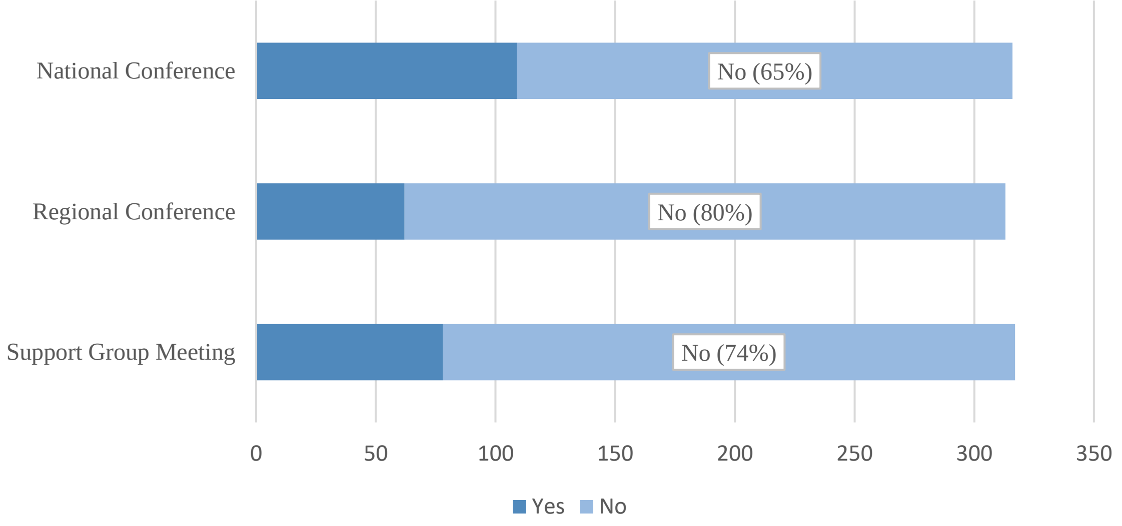


National and Regional Conferences

The majority of survey respondents had never attended the following OIF activities: National Conference (207/316, 65%), Regional Conference (251/313, 80%), Support Group Meeting (239/317, 74%). Figure 4.4 displays a summary of these responses.

Question: Have you ever attended any of the following OIF activities?

Figure 4.4



Of 316 responses, the majority (57%) reported they were not planning on attending the 2018 OIF National Conference. 25% responded that they were not sure yet of their plans.

Table 4.7 Participants of Future Activities
Question: Are you planning to attend the 2018 OIF National Conference on July 13-15 in Baltimore, Maryland? (n = 316)

Yes	58 (18.35)
No	179 (56.65)
Not sure yet	79 (25.00)

The 249 responses of the next follow-up question revealed a wide array of reasons survey participants are not planning to attend the next national conference. The standard responses include cost (51%), difficulty with traveling (28%), and the inability to get time off of work or school (18%). Seventy-three survey respondents commented with other reasons they were not planning to attend. The summary of responses to this question is outlined in Table 4.8.

Table 4.8
Question: If you are not planning to attend the next OIF National Conference, why not? (n = 249)

Cost	126 (50.60)
Too difficult to travel	70 (28.11)
Unable to get time off work/school	45 (18.07)
Other, please explain:	73 (29.32)
Comments	
Financial Reasons Need travel assistance Cost, distance, other plans Cost and unable to travel right now	
Logistical or Date Conflict Conflict with work schedule Might have close relatives from out of state visiting I am a caretaker for a disabled partner who uses a wheelchair Conflict with vacation I am self-employed and unable to take the time off Conflict with another national event at the same time period Too far away and conflicts with fun summer plans Bad time of the year to travel for us…but is on my “bucket list”	
Medical Post-partum/nursing Child with OI having surgery Just recently had a baby with OI Possible upcoming surgery	
Geographic Location/International I live in New Zealand Living outside the US Hard to travel. It would be nice to have a conference not be on the East Coast. Also having a regional conference in the Rocky Mountains Area (Denver or Salt Lake). Too far away, flights too expensive I have Type II OI, cannot fly, and it’s too far from where I live	

Too far away to feel like enough of a benefit Not in my country I live in South Africa Have two small children and husband doesn't like to travel Being from India, travel is expensive and have visa issues Can't travel easily anymore I am in India I wish I could attend Cost and travel distance, no childcare for teens with complex developmental issues Should be held in New York or a different area versus repeating MD/DC/VA
Interest My child does not want to attend the conference this year Not sure if useful Don't feel the need to go
Type of OI- Conference Content Mild OI and it seems targeted for those with more severe OI My family has mild OI, not sure of the benefit Not enough information for DI only patients My daughter has Type I OI. She's been to one national conference; however, is not interested in attending another one at this time. Not really interested, given mild OI Most of the topics deal with severe OI and that's not applicable to me I'm a grandparent of a 4 year old boy with OI/Type I Not as pertinent to my situation Feel Type I is forgotten
Connection to OI- Conference Content Children with OI are grown My son is now an adult and not interested I'm the parent of a young adult with OI so I don't think it applies to me enough to make the trip My son is deceased who had OI
Personal/Miscellaneous Having someone to attend with Anxiety Cannot be in crowds I cannot bear to see children who are suffering with OI I don't like conventions

Forty percent of 294 survey respondents felt that Medical Information Sessions were the most important aspect of attending an OIF National or Regional Conference. The second most important aspect was Peer-to-Peer Support and Encouragement (29%). Figure 4.5 displays the distribution standard responses regarding the most important aspect of attending a National or Regional Conference.

Figure 4.5

Question: In your opinion, what is the most important aspect of attending an OIF National or Regional Conference?

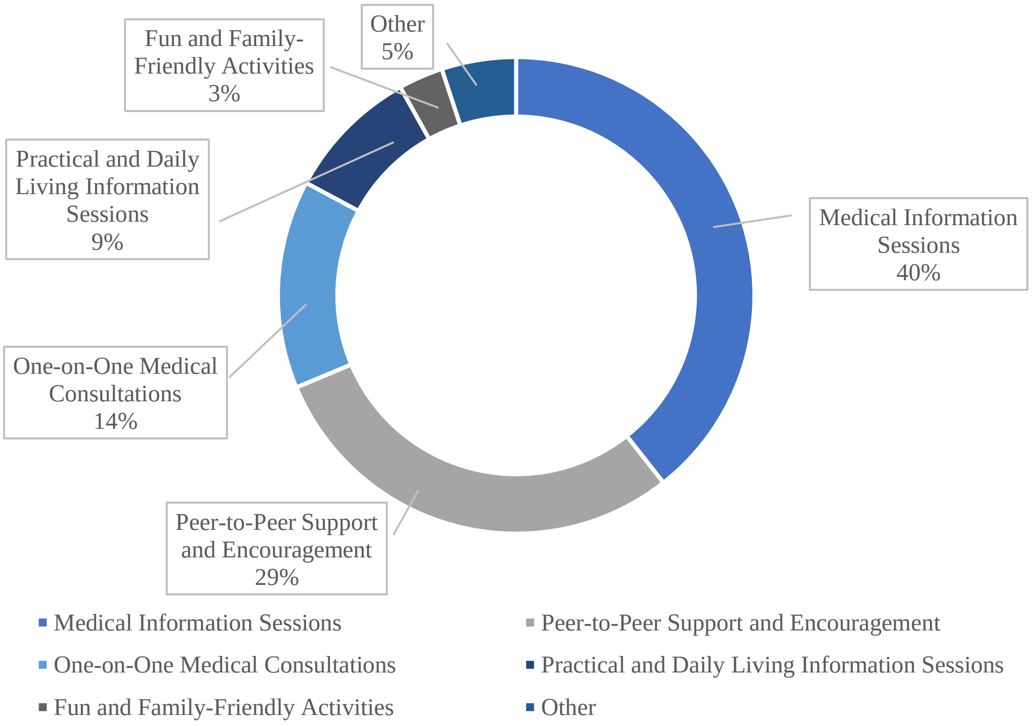


Table 4.9 outlines write-in responses of the most important aspect of attending an OIF National or Regional Conference.

Table 4.9
Question: In your opinion, what is the most important aspect of attending an OIF National or Regional Conference?

Comments
Social
See other people with the condition
Meet peers (not support related)
Multiple Reasons
Medical information is top priority for me, as is peer-to-peer for my daughter.
The interaction and discussion outside of the sessions. I have seen parents talk with adults with OI and realize the great potential of their own children. They learn about letting go and not the overprotection seen by many. People learn about their own potential as well.
All of the above, it covers all the bases and then some. You make friends for life.

A total of 150 survey respondents wrote-in suggestions for future conference session topics (95 comments) and podcasts (55). Table 4.10 organizes unique suggestions under six categories.

Table 4.10
Question: Do you have any suggestions for future conference session topics, podcasts or other activities?

Comments
Topic-Specific
Musculoskeletal
OI individuals who suffer from Costochondritis which is an inflammation of the junctions where the upper ribs join with the cartilage that holds them to the breastbone or sternum.
Spine treatments for scoliosis, kyphosis, basilar impression, etc.
Association of ligament and tendon involvement in OI. The association of arthritis due to fracture Hx.
Scoliosis treatment to improve balance
Knee/hip replacement
Re-rodding in Adults with rods protruding
Dental
More information on DI

DI only patient (not OI)

Vision

Glaucoma and thin OI-related corneas, resulting in peripheral vision loss

Pulmonary

Get the newborn OI babies in the hospitals- doctors should be more helpful with respiratory issues and possibly pushing more to get a tracheostomy. So heartbreaking and frustrating to see all these littles ones passing on. My daughter received on and it has been so great.

Physical Therapy

Practical information on how to do physical therapy with my child as she grows. Physical therapy is most important to most parents and good physical therapy is difficult to get.

Adapted exercises

Adaptive equipment to avoid stress on the body

Pharmacologic

Primers on bone biology, mobilize research support like cystic fibrosis. Activities to make OI an attractive orphan drug market

What about gene therapy research and Fresolumimab? What are dangers in participating in the trial?

Public perceptions of drug therapies

Clinical Trials, Regional differences in treatment, Meet the Scientist

Age

Complications for adults with very mild or very severe OI

Adult living with OI- how to fall

Menopause and aging- hormone replacement therapy

Pain Management and Fatigue

Impact of the opioid crisis on people with OI

More literature and best practices regarding fatigue

Psychosocial

OI/fracture-related PTSD, depression and anxiety

Mental health for those with OI and their families, i.e. parents raising children with OI, siblings at home, spouses/partners

Other

Speech

Nutrition

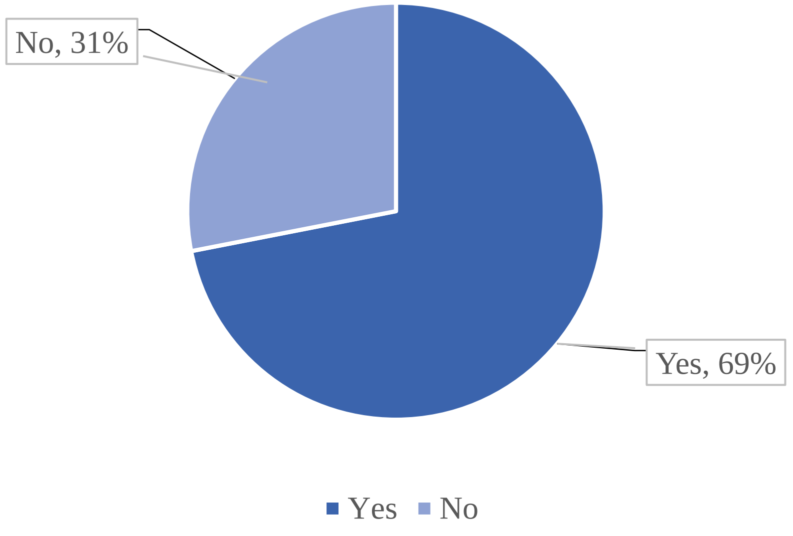
Diabetes and weight management Hydrocephalus and OI Is breastfeeding contraindicated for mothers with mild OI? Children who have OI and also have developmental delays or are on the autism spectrum Colonoscopy, hysterectomy, cancer treatments. Things for us to think about in possible future. What we should be telling our non-OI doctors who treat us.
Medical Resources OI Health and Care Clinics Transition to adult care Virtual one-on-one medical consultations
Social Services and Insurance Resources How to work with insurance companies to help them understand the need to cover adaptive equipment. Coaching on the kind of language to use on insurance appeals. Resources for child abuse scenarios- what to do when urgent action is needed How to get state health insurance to pay for aquatic therapy SSI/SSDI Insurance options after age 26
Disability Rights, Employment, School Accommodation Resources Failure to Launch Workshop, working so hard to be independent, yet fearful to get a job Disabled resources for fun, travel, vacation, cruises, etc. Continue with the Unbreakable Spirit thing you used to do. More information about 504/IEP to assist parents Vocational counseling Career choices College scholarships
Non-Medical Topics and Activities Social time for those between 21 and maybe 30- i.e. young adults More social time Information about summer camps Raising a child with OI Managing day to day life Regional sessions/discussions about medical topics, awareness support, etc. National conference should be held at a fun state where peers can go out together and do activities like swimming or wandering around at these fun places such as Las Vegas or Disneyland, Los Angeles or Hawaii. These places have many options to explore and do activities with friends like sitting at the beach or surfing.
Technological Suggestions Can you livestream conferences for those who are unable to attend? Can all podcasts and conference sessions have subtitles? Hard to hear.

Podcasts

The OI Foundation started a podcast series in 2015. The OIF currently uses this form of media to record credible and up-to-date interviews as sources of information for the constituent and medical professional communities. Most survey respondents (69%, 217/316) have never listened to or viewed an OIF podcast. The distribution is displayed in Figure 4.5. Of the respondents who have listened to a podcast, the weighted average of quality on a Likert scale of 1 (not helpful) to 5 (very informative) was 4.09. Four survey respondents commented that they did not know about OIF podcasts.

Question: Have you listened to or viewed any OIF podcasts?

Figure 4.6



Additional Characteristics and Needs of the OI Community

The weighted average of 306 responses about how comfortable they felt explaining OI to others was 4.47 on a scale of 1 (not very comfortable) to 5 (extremely comfortable). On a similar Likert scale of 1 (not very informed) to 5 (extremely informed), survey respondents on average reported their feeling of being informed about current opportunities to participate in research as 3.21. Table 4.11 displays the results to these two questions.

Table 4.11

Question: Please rate how comfortable you feel about explaining OI to others? (n = 306)	
1 = Not very comfortable 5= Extremely comfortable	
Weighted Average	4.47
Question: Please rate how informed you feel about current opportunities to participate in research about OI? (n = 305)	
1 = Not very informed 5= Extremely informed	
Weighted Average	3.21

The OI Educate Program is a new program focused on providing medically-verified information to resource contacts, or “OI educators”, who can help spread awareness and answer common inquiries through social media, phone calls, emails, support group meetings, health fairs, medical school lectures, and other activities. This program is intended to offer community members with a first line of support and not meant to provide medical advice. It has been advertised once in the OIF e-newsletter and print newsletter Breakthrough. Despite the potential lack of community awareness about this new initiative, the majority (55%) of 300 respondents were interested in participating in the OI Educate program.

Thirty-nine percent (118) of 305 respondents were interested in volunteering to assist with a local support group. Nine percent (26) noted that they already volunteer with a local support group. Twenty-three percent (69) of 304 respondents marked that they were interested in coordinating a local activity or fundraiser for 2018 National OI Awareness Week or Wishbone Day. The results to this question are outlined in Figure 4.7.

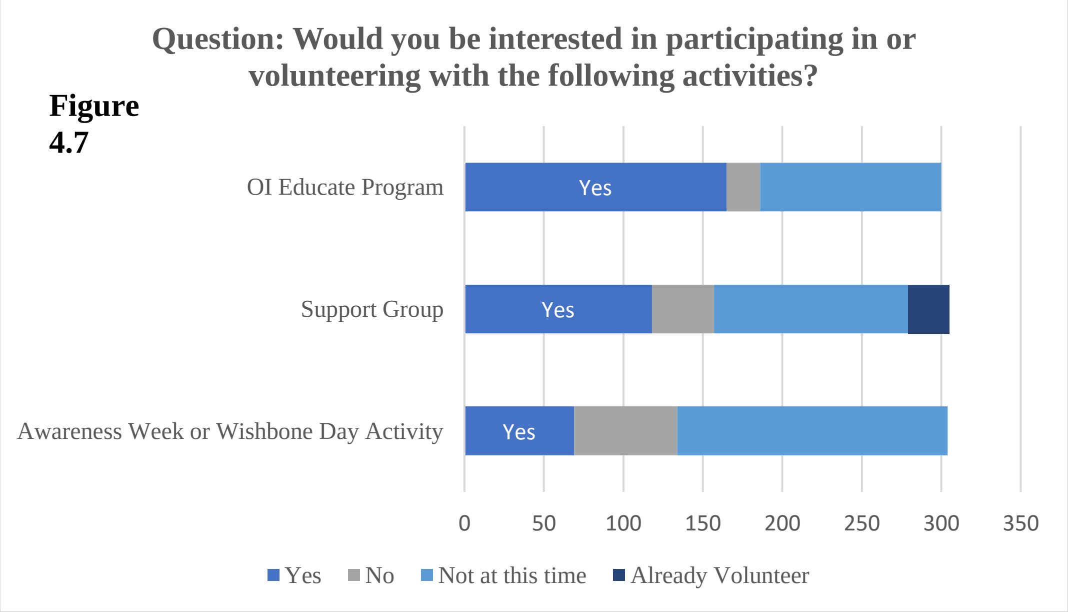


Figure 4.7

Preferred Formats to Access Information about OI and the OI Foundation

Sixty-nine percent of 305 respondents identified the OIF email newsletter as their most preferred format to access information about OI and the OIF. The second most preferred format (59%) was the OIF website, followed by Facebook (43%), and the OIF Print Newsletter (Breakthrough) (30%). The complete distribution of responses is shown in Figure 4.8 and Table 4.12.

Question: Which format(s) do you prefer accessing information about OI and the OIF? (Please select all that apply.)

Figure 4.8

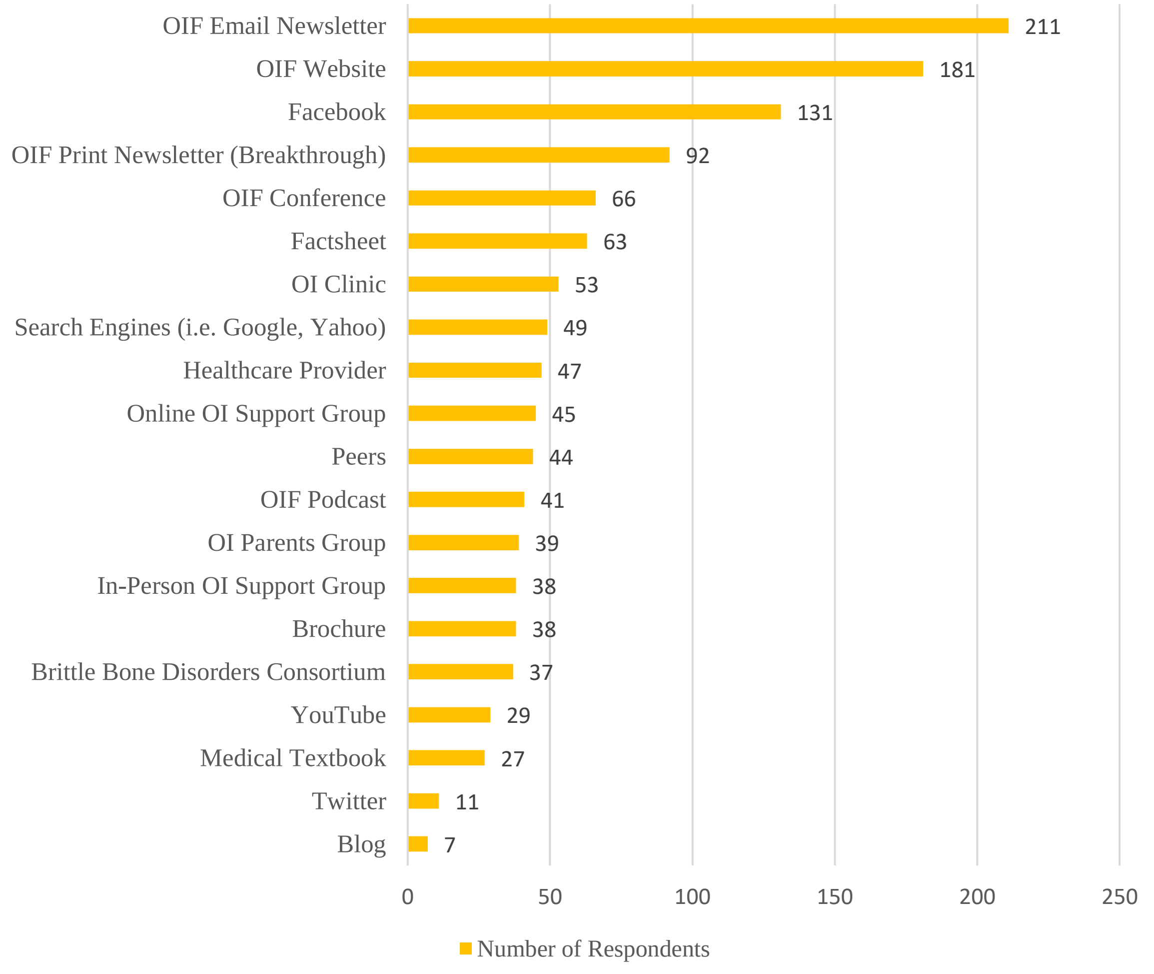


Table 4.12
Question: Which format(s) do you prefer accessing information about OI and the OIF? (Please select all that apply.)

Formats	**n (%)**
OIF Email Newsletter	211 (69.18)
OIF Website	181 (59.34)
Facebook	131 (42.95)
OIF Print Newsletter (Breakthrough)	92 (30.16)
OIF Conference	66 (21.64)
Factsheet	63 (20.66)
OI Clinic	53 (17.38)
Search Engines (i.e. Google, Yahoo)	49 (16.07)
Health Provider	47 (15.41)
Online OI Support Forum	45 (14.75)
Peers	44 (14.43)
OIF Podcast	41 (13.44)
OI Parents Group	39 (12.79)
In-Person OI Support Group	38 (12.46)
Brochure	37 (12.46)
Brittle Bone Disorders Consortium	38 (12.13)
YouTube	29 (9.51)
Medical Textbook	27 (8.85)
Twitter	11 (3.61)
Blog	7 (2.30)
Other (please specify)	6 (1.97)

Two write-in responses of note include “PubMed” as a preferred format for information, as well as “Former OIF Program Services Director Mary Beth, who was a wealth of information and assistance to me.”

As a service to the OI Foundation, the survey tool asked respondents to list primary care physicians or specialists who they would like to ensure are listed in the OIF Physician Referral Database. Fifty six respondents commented with names of physicians and clinics or hospitals. To preserve anonymity and confidentiality, the data was excluded from the analysis.

Lastly, the OIF Program Services Survey concluded with a text box to share any additional comments or suggestions. Table 4.12 lists closing quotes from survey respondents.

Table 4.13
Question: Please share any additional comments or suggestions.

Comments

Positive Feedback

- Thank you for working hard to end OI.
- Thanks for all that you do!
- I am doing a clinical trial at the moment and it’s all because of the information I read on **www.oif.org**, so thank you!
- There is limited information about OI in my country. I find the newsletters very helpful.
- Thanks for all you do! Expand services and clinics to the southeast. Offer more training, fellowships, and internships for young doctors in various areas of the country. Also, I don’t know what the OI Educate Program is.
- OIF needs a mentor program. We need to be utilizing adults with OI as a resource for parents of children with OI. Adults have been a true blessing on this journey. They are our greatest asset.
- I am grateful for this organization and its current information on my bone disease. It is information I give to my doctor.
- The quality and quantity of information the OIF has provided since I was first aware of the website is amazing. It is very helpful and very much appreciate- especially for those of us who live in other countries.
- You all are doing great!
- Please keep OIF information coming.

Information and Support Opportunities

- More support for aging OI population
- Make conference available online
- My siblings would like to have more information for adult siblings
- We are new to OI. Our new grandson was just diagnosed two weeks ago. Trying to gather as much information as we can.
- It would be very useful if there was a checklist of which doctors ought to see at different intervals for routine care. If it could be updated annually/every few years and emailed out. My quick list includes audiology, ophthalmology, dentistry, pulmonology, gynecology, cardiology, endocrinology, neurology, orthopedics, and a regular physical. Then we’d all complain even more about being professional patients, but at least people would know what is considered standard.
- I started busting tendons at age 50. I always broke bones. Never hear anything about that.
- I will be 78 years of age soon. I live alone and my energy level is very low. I am unable to travel, but would appreciate any opportunity to learn.

I do not like to travel. I get achy in the car and I will not fly. I wish there was an OI support group where I live. I am a positive person and my adult son has a positive outlook also. I have a lot of experience but no one to share it with.
Research Suggestions
OIF needs to market for therapeutic development. CF has a great model to follow. Advocate and participate in developing a cure!
Would be great to have BBD Consortium information spread out to the community.
Physician Referral and Training Suggestions
List of doctors from foreign countries should be included on website
It’s very difficult to find experienced doctors in my area
I am in a medical desert- more training for physicians
I hope more physicians can be recruited for the physician referral database. Many areas have none.

Further Analysis

Cross-tabulation analysis revealed further insights about the overall highest ranked informational topics based on the survey respondents’ type of OI, or the type of OI of their connection. The topics of bone/orthopedic issues ranked highest among community members affected by moderate OI (71%). Both pain management and adult general health (55%) ranked highest among individuals who were not sure what type of OI they or their connection have. Hearing loss ranked highest (41%) for individuals with mild OI. See Figure 4.9 for the complete chart.

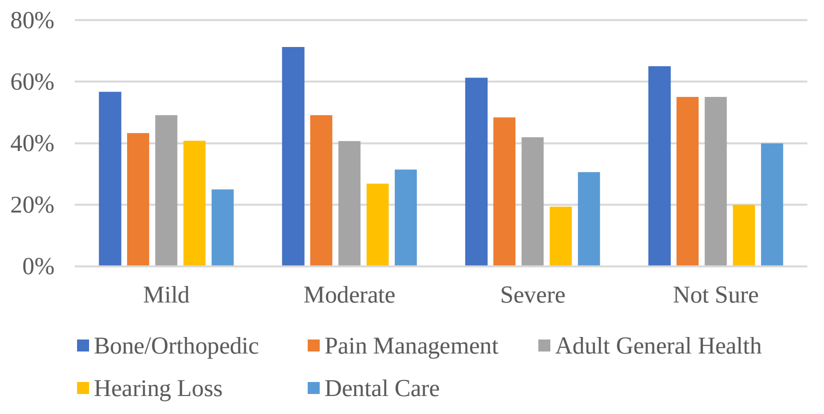


Figure 4.9 **Top 5 Topics by Type of OI**

The overall highest ranked informational topics stratified by age group revealed that the need for information on bone/orthopedic issues decreases for survey respondents older than 54 years old. This cross-tabulation also reveals that the need for information about pain management and hearing loss generally increase with age, and the need for information about dental care is consistent across all age groups. See Figure 4.10 for the full chart.

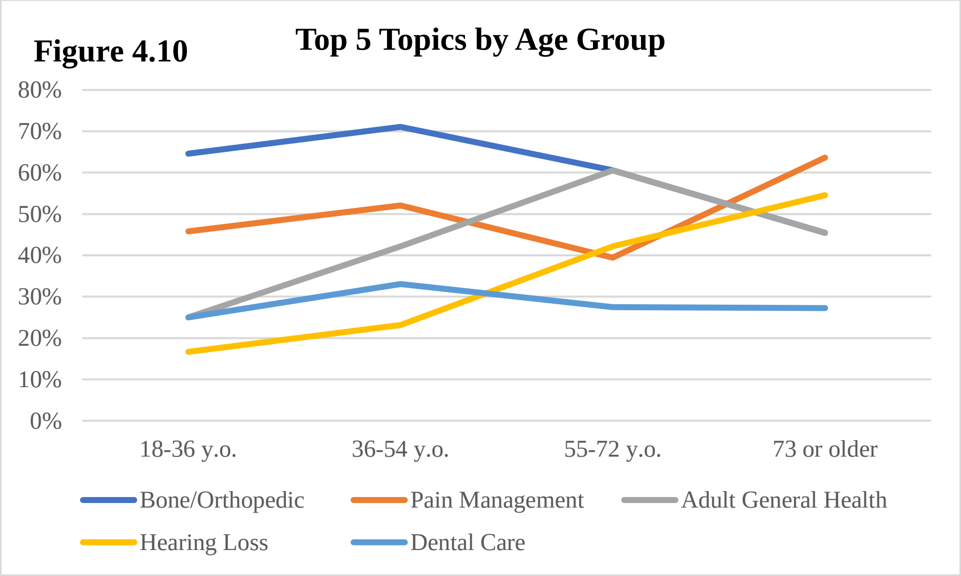


Figure 4.10

Figure 4.11 reveals positive slopes for 2 of the top 5 preferred formats (OIF Email Newsletter and OIF Print Newsletter Breakthrough) for information as age increases. See Figure 4.11 for complete distribution of preferred format rankings by age group.

Figure 4.11 **Top 5 Preferred Formats by Age Group**

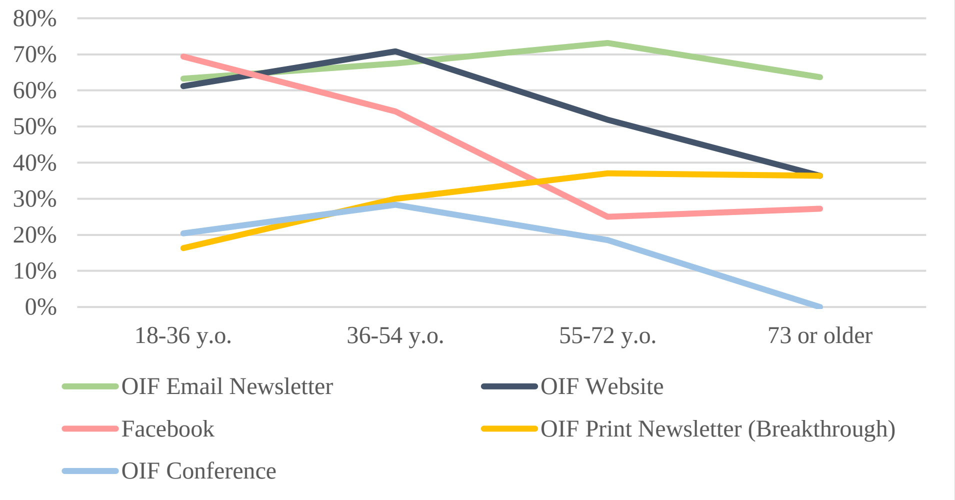
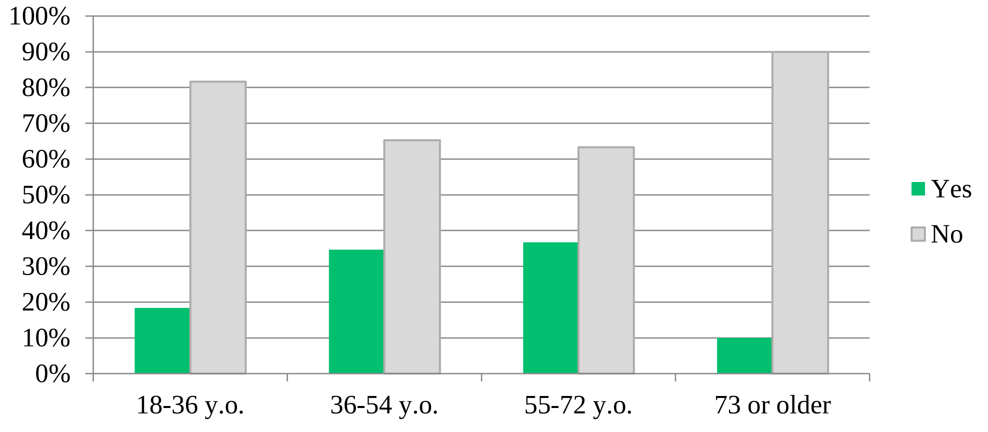


Figure 4.12 confirms that the majority of survey respondents are either not aware or not experienced with OIF Podcasts. Thirty-seven percent of survey respondents in the age group of 55-72 years old reported that they have listened to or viewed an OIF Podcast.

Figure 4.12 **OIF Podcast Usage by Age Group**



Chapter 5: Discussion

The results of the OIF Program Services Survey demonstrate a cost-efficient approach to implementing evidence-based patient education materials and programs. The number of total responses (385), survey completion rate (77%), and number of write-in responses collected over a three-week period indicate a highly engaged and motivated constituent base. The five highest ranked topics for informational materials and resources varied when stratified by age group, gender, and connection to OI. Print and web-based materials are well-utilized, ranking high as preferred formats for OI-related information and resources. The reach and representation of the survey, as shown through the range of international and geographically dispersed participants, aligns with trends of many other OI-focused surveys, such as the OI-Adult Natural History Initiative (OI-ANHI) study (Tosi et al., 2015). Tosi's 2010 study documented a similar trend that researchers of the current study observed, which includes "underrepresentation of the male gender and non-Caucasian race" (Tosi et al., 2015).

One of the positive outcomes of this study was the identification of "new" or prominent subpopulations within the OI community. The needs of the population affected by mild OI was already known by the OI Foundation, as it is well-established that OI is an extremely heterogeneous disorder; however, the overwhelmingly large response requesting more information tailored for less severe types of OI was clear throughout the survey results. Table 4.2 shows that "Managing Mild OI" was ranked by almost 1/3 of survey respondents, and tied for the sixth most important topic for OI-related informational materials and resources. This category could be further divided into several subcategories such as aging with mild OI or hearing with mild OI on future informational needs assessments.

One of the emerging subpopulations noted in the survey analysis is the parent population of grown children living with OI. Table 4.8 documents several comments from parents who felt that conference sessions did not apply to them because their children are now grown. In the past, the OI Foundation successfully organized "peer-to-peer discussion roundtables" for smaller subpopulations, such as grandparents or siblings of individuals living with OI. This could be a solution to address the needs of emerging subpopulations. The recognition of various subpopulations regarding the type of OI, age, and geographic location reinforces the idea of regional conferences and special topic workshops. The geographic diversity of respondents and comments about cost and transportation related challenges validate the need to implement more regional conferences, webinars and virtual activities.

The responses about preferred formats of accessing information about OI and the OIF were not expected. When designing the survey, members of the study team hypothesized that Facebook or another social media would rank as the most preferred format to receive information. The OIF email newsletter was ranked by 69% (211) of survey respondents as their most preferred format, followed by the OIF website (59%). While the OIF endeavors to continue expanding webinars, podcasts and more non-traditional modes of disseminating information, other established communication vehicles are valuable and should not be abandoned at this point. These results have significant implications on the communication strategies of the OIF.

Strengths

A major strength of this study was the response rate over a period of three weeks. The ability to access quick feedback is valuable for many purposes. The survey helped create awareness around newer initiatives, such as podcasts and the OI Educate Program. One survey respondent indicated in the last question to share any additional comments (Table 4.13) that they were not aware of the OI Educate Program; however, they were still interested in learning more. This study was successful in collecting data points on a wide range of topics, programs and publications, while providing a baseline structure and tool for future assessments.

Limitations

This survey was not designed with validated measures; therefore, it may not be reliable or replicable with another set of survey respondents within the OI population. There were limited published examples of needs assessment surveys in disease-specific and rare disease populations. Of the literature available on related topics, there were sporadic reports not published in academic journals or developed with IRB approval. Two members of the study team, including the Principal Investigator, are employed by the OI Foundation. The study team was careful to reduce the potential risk of bias.

An often reoccurring theme in research is that women typically far outnumber men as respondents to surveys (Tosi et al., 2015). Though not surprising, more outreach to male community members is needed to increase their current 15% response rate to closer towards equally representative (50%). The overall response and completion rate was high; however, many survey respondents skipped questions, resulting in 23% incomplete survey responses.

Future Directions

The OIF Program Services Survey helped identify new topics and categories to implement in programs and publications. During the period of data collection and analysis, several inquiries from other rare disease organizations and international counterparts to the OIF requested the survey template. One of the goals of this project is to provide an effective survey instrument to share with other patient advocacy groups. Once another round of feedback from OIF and BBDC leadership is implemented and the manuscript is submitted, the template will be shared with other groups. The OIF would also like to implement an annual or biennial Program Services Survey as a shared international resource and recruit participants from more sources.

Chapter 6: Population and Public Health Implications

As the science of health education and patient engagement evolves, data on health information consumers benefits the patient population in a variety of contexts. With the use of needs assessments, more effective methods of communication are identified for physicians and patient advocacy groups, and the constituent community and patient population are able to voice their most critical needs and concerns. The results of this survey reiterate the findings from a study published in the Journal of Pediatric Nursing, which emphasized the importance of "understanding social contexts, including families, peers, everyday activities, and relationships" in patient education and the development of informational materials (Driessnack, 2009). The OI Foundation is dedicated to building powerful partnerships between patients, scientists, and physicians and empowering a well-informed and "research ready" community.

Lessons in Strategic Health Communications

Setting priorities for programs and publications can be challenging for small patient advocacy groups with limited resources. Non-profits focused on rare disorders must plan and invest strategically in their program services to maximize the impact for all constituent communities. Results from the OIF Program Services Survey provided valuable recommendations to improve services and ensure that activities are well-aligned with the missions of the OI Foundation and the Brittle Bone Disorders Consortium. The survey results had an immediate effect on the design of upcoming communications and programs. Studies have shown that personal preferences on formats and timing of information may affect an individual's ability to retain and process information and their ability to engage with their care team, which could ultimately influence health outcomes (Jessup et al., 2016). Considerations must be made for the large variety of preferences in information formats, as shown by the survey results. It is critical that publications and programs are easy for patients to access, as difficulty could exacerbate disparities for populations in the most need of information and support. The OIF Program Services Survey confirmed the critical need for increased collaboration, effective communication, and forceful advocacy amongst all stakeholders (Hart et al., 2014).

One of the OI Foundation’s tagline phrases, as seen in the collage of logos in Figure 6.1, is Unbreakable Spirit®. Trademarked by the OIF in 2005, the slogan is commonly referred to in the OI community as a sentiment that people living with OI are much more than their fragile skeletons. The “Unbreakable Spirit” slogan evolved into a theme that is expressed in OI communities worldwide. The strength and spirit of the OI population was displayed in the results of the OIF Program Services Survey. Many respondents suggested more mentorship programming and peer-led discussion groups on specific topics. Based on the quick response rate, OIF constituents seemed eager to contribute their ideas and participate in a study that could possibly benefit them or their families in the near future.

Figure 6.1 OIF Logos and Tagline

Conclusion- Empowering a "Research-Ready" Community

"The reward for work well done is the opportunity to do more." – Jonas Salk, MD

In summary, a critical component to the future success and impact of the OI Foundation is the involvement of volunteer leaders in the research and medical community (Hart et al., 2014). At the 2018 National Institutes of Health Rare Disease Day Celebration, Margie Frazier from the Batten Disease Support and Research Association emphasized the importance of collaboration. Adapted in Figure 6.2, she illustrated funders, scientists/physicians/industry, and patients as the major stakeholders or "gears" in drug development and research. She concluded with an image of an engineering metaphor similar to Figure 6.2. She stated, "Our role is to be the grease that helps the gears move. That involves communication, collaboration, hope, energy. We're happy to be the grease."

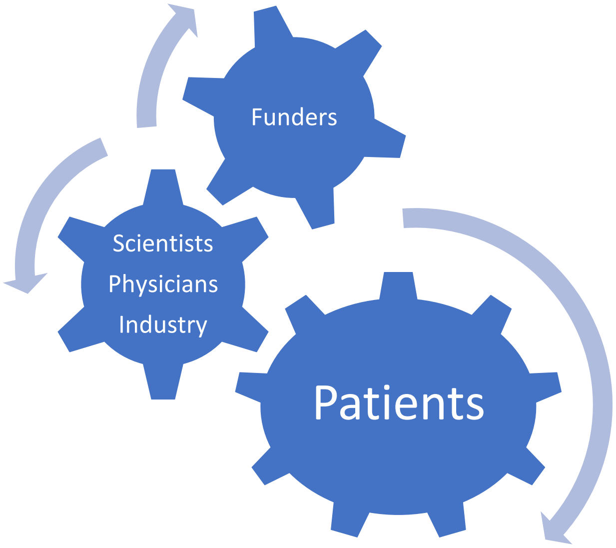


Figure 6.2 Adapted Graphic from the Batten Disease Support and Research Association

Informational needs assessments and health literacy tools assist in understanding the dynamic needs of a patient population and larger constituent community. The OI Foundation serves as an advocate to expand the understanding of the causes and consequences of OI, and acts as a conduit to accelerate research for improved treatments and eventually a cure. As OI-related educational programs and services continue to reach new and larger populations, the OI Foundation will continue to build on the history of nearly five decades of mission-driven work and partnerships.

Chapter 7: Journal Article

Information Needs and Patient Engagement of Individuals affected by Osteogenesis Imperfecta

Patient Education and Counseling Journal

Corresponding Author: Petra Harvey, petra.harvey@gmail.com

Iris Smith, PhD
Emory Rollins School of Public Health

Tracy Hart, CEO
Osteogenesis Imperfecta Foundation

Reid Sutton, MD
Baylor College of Medicine
Brittle Bone Disorders Consortium

Kara Ayers, PhD
University of Cincinnati University Center for Excellence in Developmental Disabilities

Osteogenesis Imperfecta Foundation
804 West Diamond Avenue, #210
Gaithersburg, Maryland 20878

Petra Keturah Harvey

Identifying Informational Needs of Individuals affected by Osteogenesis Imperfecta and Bridging the Gap to Increase Health Literacy and Patient Engagement

Abstract

Introduction: Populations affected by rare genetic disorders such osteogenesis imperfecta (OI) often have complex health information needs yet limited resources for timely and accurate information about their condition. It is vital that patient advocacy groups and healthcare providers understand the dynamic informational needs of individuals living with OI and their support networks.

Objective: The study accessed informational needs within the OI community in order to prioritize future OI-related programs and publications.

Methods: A needs assessment survey tool was developed and disseminated by the OI Foundation. Ethics approval was secured. Quantitative and qualitative data were collected over a three-week period.

Results: The five highest rated topics for OI-related informational materials and resources include bone/orthopedic (65%), pain management (46%), adult general health (45%), hearing loss (30%), and dental care (30%). Sixty-nine percent of 305 question respondents identified the OIF email newsletter as their most preferred format to access information about OI and the OIF. The second most preferred format (59%) was the OIF website, followed by Facebook (43%).

Discussion: The number of total responses (385), survey completion rate (77%), and number of write-in responses collected over a three-week period indicate a highly engaged and motivated constituent base. The five highest ranked topics for informational materials and resources varied when stratified by age group, gender, and connection to OI. Print and web-based materials are well-utilized, ranking high as preferred formats for OI-related information and resources. While the OIF endeavors to continue expanding webinars, podcasts and more non-traditional modes of disseminating information, other established communication vehicles are valuable and should not be abandoned at this point. The results of the OIF Program Services Survey demonstrate a cost-efficient approach to implementing evidence-based patient education materials and programs.

Introduction

In the modern culture of information overload, navigating the complex healthcare system can require a tremendous amount of skill and persistence. Many times patients must piece together information relevant to their condition from a variety of sources, including their physician team, medical brochures or textbooks, support groups, social networking sites, and search engines. The overwhelming amount of general consumer health information available online for the general population can make the task of accessing accurate information difficult. Retrieving, processing, and applying information about a rare health disorder like osteogenesis imperfecta (OI), also known as brittle bone disease, can be a confusing task in the digital era. Twenty years ago, a spokesperson for the US Department of Health and Human Services advised, “Trying to get information from the Internet is like drinking from a firehose, and you don't even know what the source of the water is” (McLellan, 1998). While online healthcare consumers may have physical access through various online sources, the use of jargon and highly technical language can also make information inaccessible. Addressing unmet informational and supportive resource needs is necessary to increase health literacy and patient engagement, and ultimately improve patients’ quality of life and overall health outcomes.

Several diagnostic tools have been developed and validated to identify patients with limited health literacy. Two of the most widely used measures include the Test of Functional Health Literacy in Adults (TOFLA) and Rapid Estimate of Adult Literacy in Medicine (REALM) (Nutbeam, 2009). Developed in 1994, the TOFLA measures adult’s functional literacy levels in relation to numeracy and reading comprehension of real patient education materials (Parker et al., 1995). The REALM is a similar standardized test that uses common words in word recognition questions, and can be adjusted based on the age and language of the primary target population (Parker et al., 1995). Teach-back or “show-me” methods are common approaches that physicians employ on an individual level to confirm understanding of health information (Williams et al., 2002). Recent research studies have begun to criticize the use of health literacy assessments, emphasizing the need for “more practical and efficient approaches to produce more accessible and easier-to-understand materials” instead of determining new ways to test health literacy (Egbert et al., 2009).

Individuals affected by OI are typically encouraged to connect with the Osteogenesis Imperfecta Foundation (OIF), whose mission is to improve the quality of life for people affected by OI through research to find treatments and a cure, education, awareness, and mutual support. Established in 1970, the OI Foundation has served as a hub for information for the estimated 50,000 children and adults nationwide that live with OI, and the larger community (Hart et al., 2014). Nearly fifty years of offering educational resources and programs tailored to the OI community have been helpful in navigating individual complex decisions, empowering the community, and preparing patients to engage in clinical research.

Common sources of social support and health information organized by the OI Foundation include family conferences and support groups, factsheets, newsletters, podcasts, webinars, and virtual interactive forums. Throughout nearly five decades of service, the OI Foundation “became the first stop for people looking for information on OI because of a new diagnosis, because of a new symptom, or because of a new challenge they are facing” (Hart et al., 2014). Staying in constant communication with constituents gives the OIF unique and valuable insight into the informational needs and sources that the community relies on.

Driven by its mission, the OIF is not only committed to fulfilling programs focused on educational and mutual support, but it also strongly supports research that accelerates a better understanding of OI and improved treatments. The OIF established the OI Adult Natural History Initiative (OI-ANHI) in 2010 to “give voice to the health concerns of the OI community” (Tosi et al., 2015). Led by Dr. Laura Tosi, a long-time member of the OIF’s Medical Advisory Council, this research study collected “self-reported disease severity, reported symptoms and health conditions, estimated the impact of [the] concerns on present and future health-related quality of life (QoL) and completed a Patient-Reported Outcomes Measurement Information System (PROMIS®) survey of health issues” (Tosi et al., 2015). Patient-centered outcome research is necessary to produce engaged patient populations and promote the partnership between medical professionals and patients. With the goal of discovering effective therapies and improving health outcomes, it is the objective of healthcare professionals and patient advocacy groups like the OI Foundation to bridge the gaps in knowledge and prepare patients to make informed decisions about their healthcare and future research.

Methods

Survey Instrument Design

Based on evidence from published needs assessments in other disease-specific populations, a twenty-seven question Program Services Survey was developed for the osteogenesis imperfecta community (Anand et al., 2013; Hamilton et al., 2015). A mix of multiple-choice, check box, and Likert rating scale questions were implemented. The target audience of the survey instrument included OIF constituents and individuals over 18 years of age who are affected by OI. Feedback from the OIF's Board of Directors and Medical Advisory Council were incorporated in the content and design. The suggested categories of sociodemographic data included sex, age range, connection to OI, and the type of OI (self or connection). Race, ethnicity, geographic location, education background, and income level were sociographic categories not included to preserve anonymity. The survey template and introduction letter were printed in lay language and mirrored on the Survey Monkey platform for online access.

Ethics Consideration

This study had minimal risks, and was declared exempt by the Emory University Institutional Review Board on January 30, 2018 (Appendix C). Informed consent was prompted for online and paper survey respondents before questions were presented, and participants were assured of anonymity and confidentiality.

Recruitment and Data Collection

Participant recruitment activities included an email announcement (Appendix D), mailed paper surveys (Appendix B), and social media announcements (Appendix E) with instructions on how to complete the survey. Four reminders were disseminated through email and social media (Appendix E). The survey was administered via both the Survey Monkey platform and a mailed paper form. Data was collected over a three week period in February 2018.

Data Analysis

The target goal for survey participation was 300. Of the 400 online and paper survey responses collected over three weeks, 15 respondents indicated they did not wish to be counted in the analysis, leaving a total of 385 responses. Frequency and cross-tabulation statistics were generated for raw quantitative data. Write-in comments and responses were analyzed and coded according to themes to avoid redundancy.

Results

A total number of 385 participant responses were evaluated from the OIF Program Services Survey. The survey had a 77% completion rate and took online respondents an average of five minutes to complete. Respondents were permitted to skip responses to any question, so the number of responses (n) varies for each question or category.

Most participants (84%) who completed the survey were women. The largest age category reported among survey respondents (40%) was ages 36-54 years old, followed by 55-72 year old respondents, who represented 38%. The majority of respondents (58%) reported that they had OI themselves. Forty-four percent of respondents reported they were parents of children or adults with OI. The remaining categories of respondents' connections to OI varied: 14% were family members of an individual with OI, 8% were grandparents of an individual with OI, 7% were health professionals, 4% were spouses married to a person with OI, 1% of respondents were friends of an individual with OI. The categories of respondent's connections to OI were presented in a check-box response question, so respondents could select all that applied.

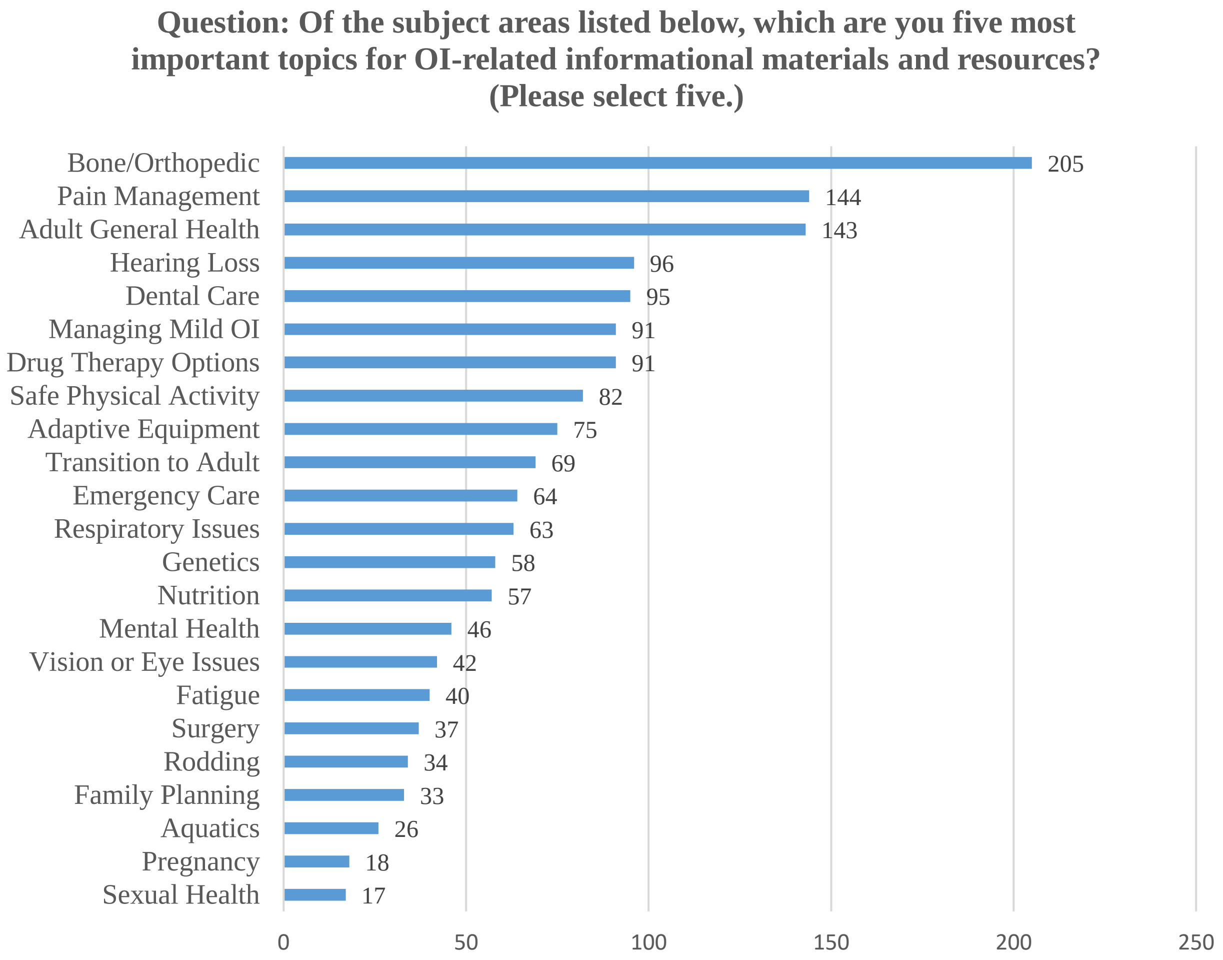
The full analyses of demographics collected on this survey are outlined in Table 1.

Table 1
Survey Respondent Demographics (overall n = 385)

Category	**n (%)**
Sex	**309**
Male	45 (14.56)
Female	260 (84.14)
Prefer Not to Disclose	4 (1.30)
Age	**307**
18-36 years old	50 (16.29)
36-54 years old	124 (40.39)
55-72 years old	116 (37.79)
73 or older	13 (4.23)
Prefer Not to Disclose	4 (1.30)
Connection to OI	**308**
Self	179 (58.12)
Parent/Guardian	134 (43.51)
Spouse	11 (3.57)
Grandparent	26 (8.44)
Family Member	44 (14.29)
Friend	4 (1.30)
Health Professional	21 (6.82)
Other	16 (5.20)
Type of OI (Respondent or Connection)	**307**
Mild	125 (40.72)
Moderate	109 (35.50)
Severe	69 (22.48)
Not Sure	23 (7.49)
Not Applicable	7 (2.28)
Prefer Not to Disclose	1 (0.33)

The five highest rated topics for OI-related informational materials and resources included: bone/orthopedic (65%), pain management (46%), adult general health (45%), hearing loss (30%), and dental care (30%). Figure 1 shows the most important subject areas by topic and their corresponding responses.

Figure 1



Question: Of the subject areas listed below, which are you five most important topics for OI-related informational materials and resources? (Please select five.)

On a Likert scale of 1 (not helpful) to 5 (very informative), survey participants were asked to rate the quality of OIF materials and resources. 119 of 305 respondents (39%) found OIF materials and resources very informative. The weighted average of 305 responses was 4.04/5. The breakdown of responses is displayed in Figure 2.

Question: Please rate the quality of OIF materials and resources.

Figure 2

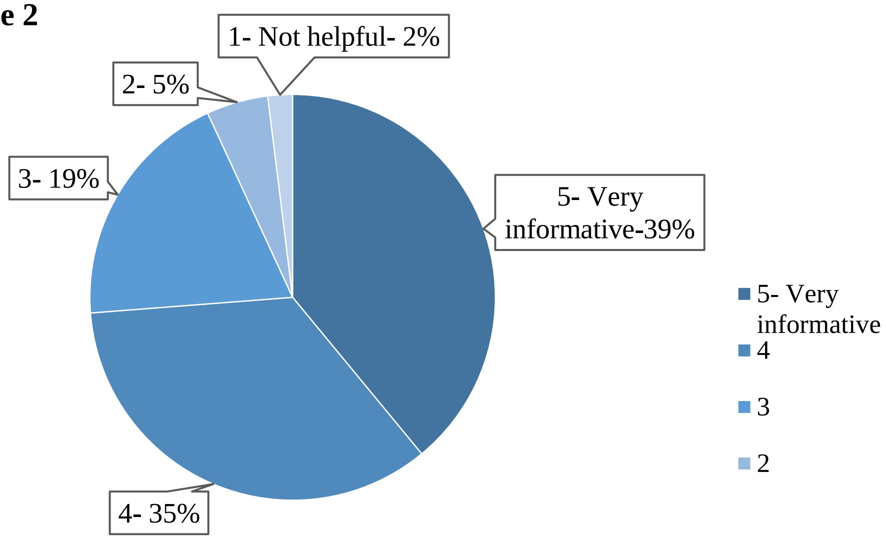
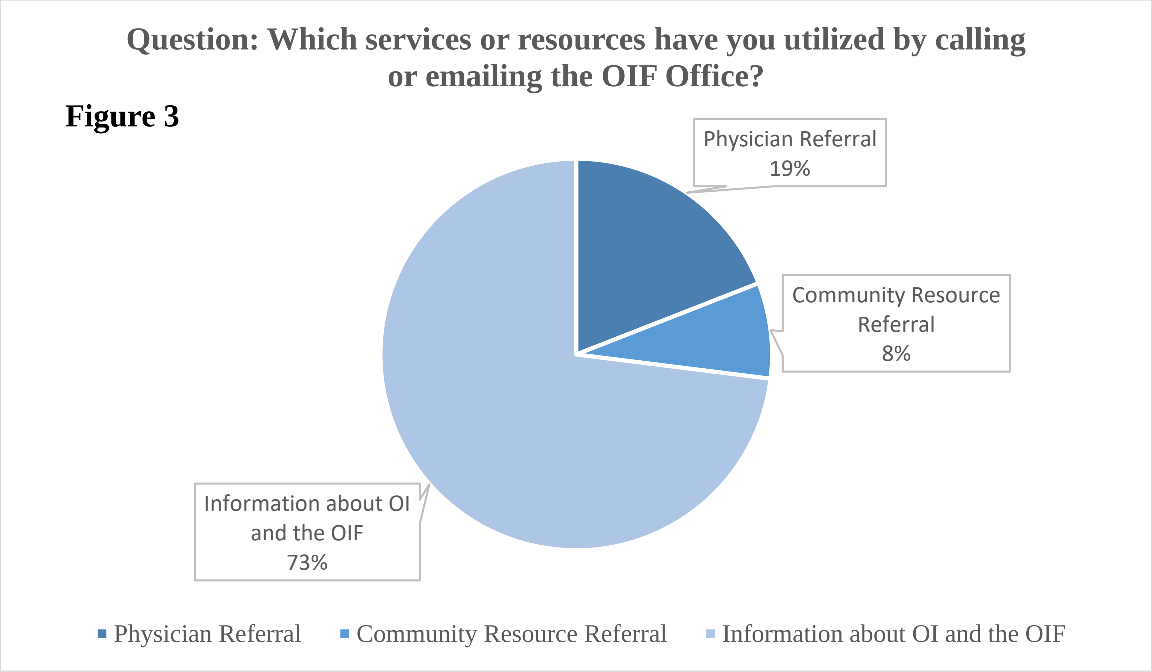


Figure 3 summarizes the proportion of respondents who have utilized OIF Information Center services or resource by calling or emailing the OIF Office. Of the 234 survey participants who responded to this question, 73% reported they had received information about OI and the OIF, 19% reported they have received a physician referral, and 8% reported they have received a community resource referral.

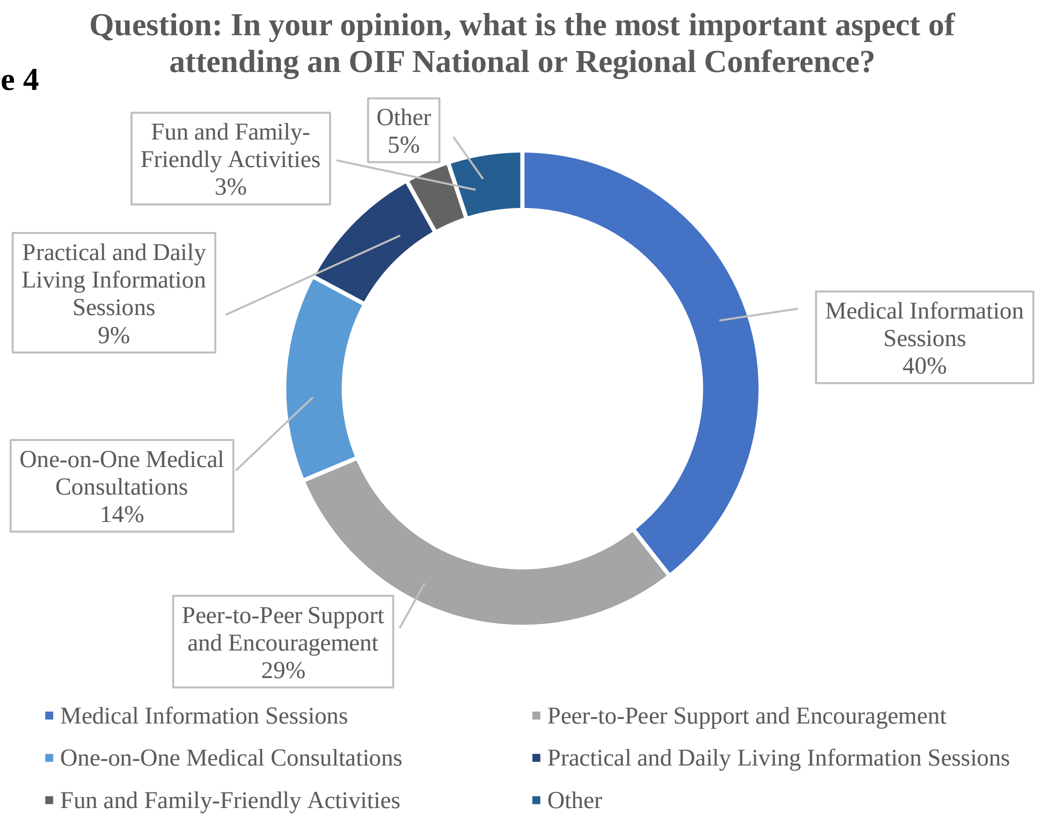


Question: Which services or resources have you utilized by calling or emailing the OIF Office?

Figure 3

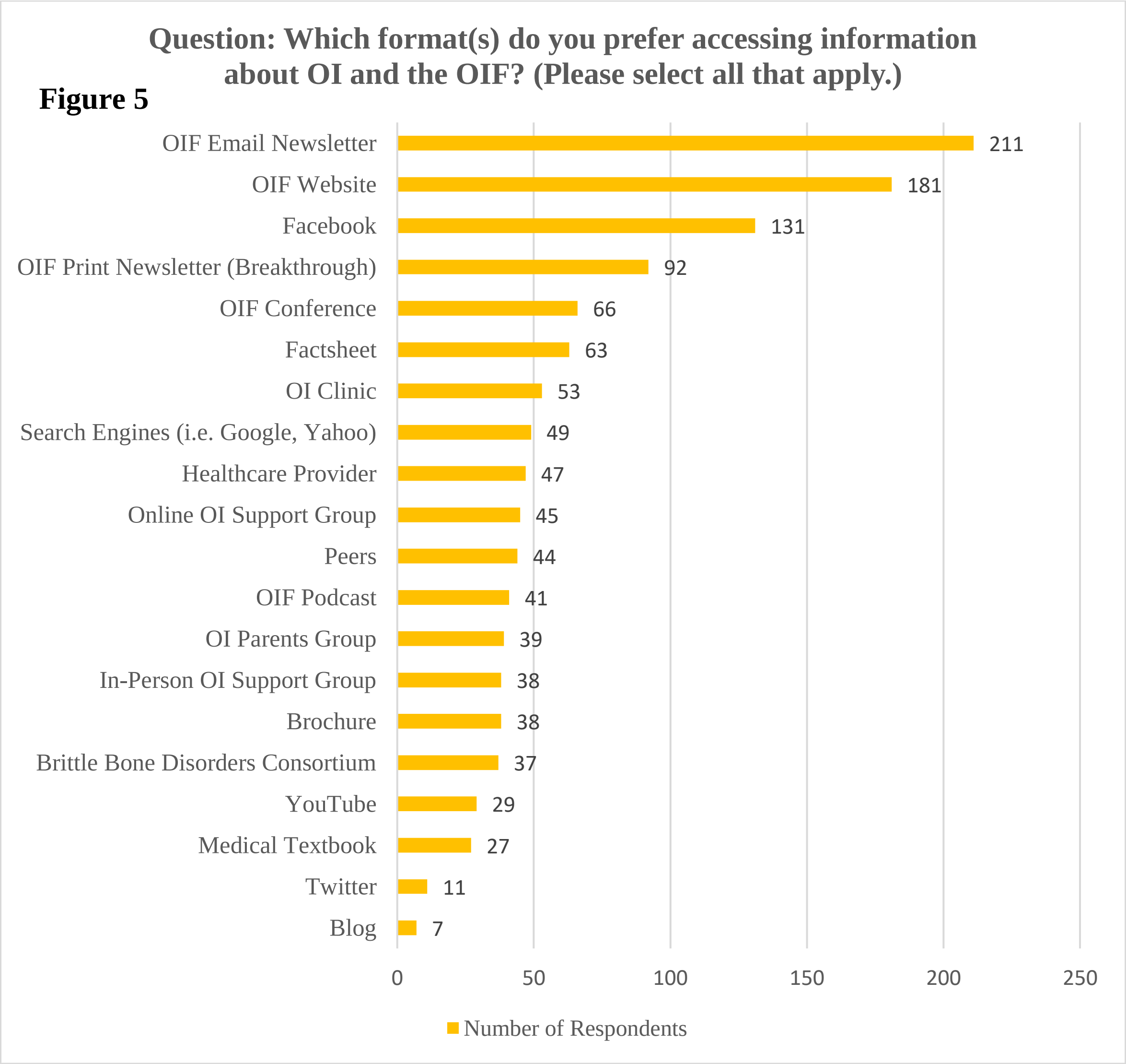
Forty percent of 294 survey respondents felt that Medical Information Sessions were the most important aspect of attending an OIF National or Regional Conference. The second most important aspect was Peer-to-Peer Support and Encouragement (29%). Figure 4 displays the distribution standard responses regarding the most important aspect of attending a National or Regional Conference.

Figure 4



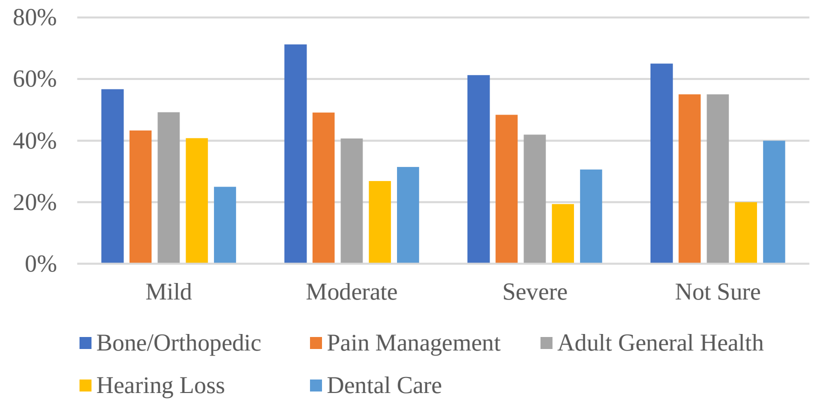
Sixty-nine percent of 305 respondents identified the OIF email newsletter as their most preferred format to access information about OI and the OIF. The second most preferred format (59%) was the OIF website, followed by Facebook (43%), and the OIF Print Newsletter (Breakthrough) (30%). The complete distribution of responses is shown in Figure 5.

Figure 5



Cross-tabulation analysis revealed further insights about the overall highest ranked informational topics based on the survey respondents' type of OI, or the type of OI of their connection. The topics of bone/orthopedic issues ranked highest among community members affected by moderate OI (71%). Both pain management and adult general health (55%) ranked highest among individuals who were not sure what type of OI they or their connection have. Hearing loss ranked highest (41%) for individuals with mild OI. See Figure 6 for the complete chart.

Figure 6 **Top 5 Topics by Type of OI**



The overall highest ranked informational topics stratified by age group revealed that the need for information on bone/orthopedic issues decreases for survey respondents older than 54 years old. This cross-tabulation also reveals that the need for information about pain management and hearing loss generally increase with age, and the need for information about dental care is consistent across all age groups. See Figure 7 for the full chart.

Figure 7

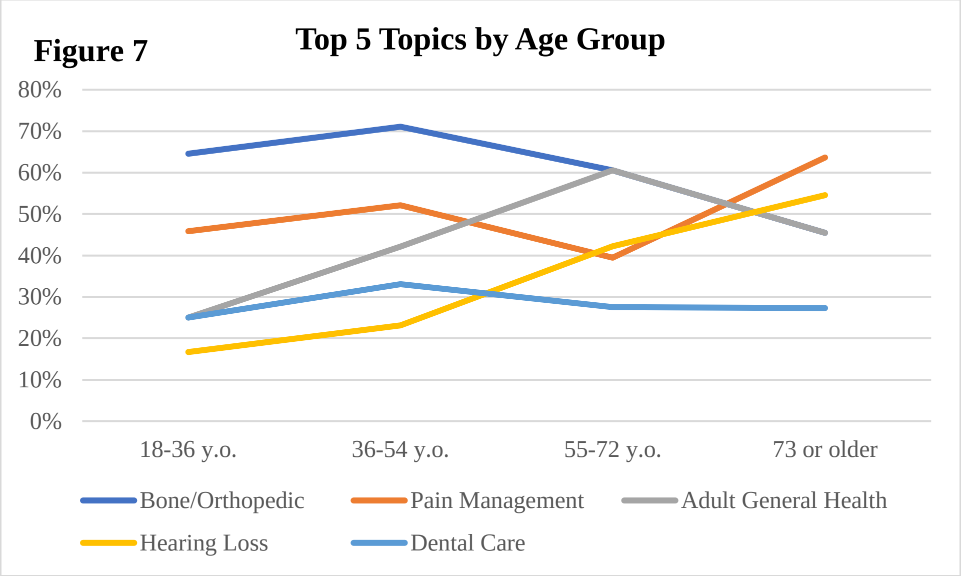
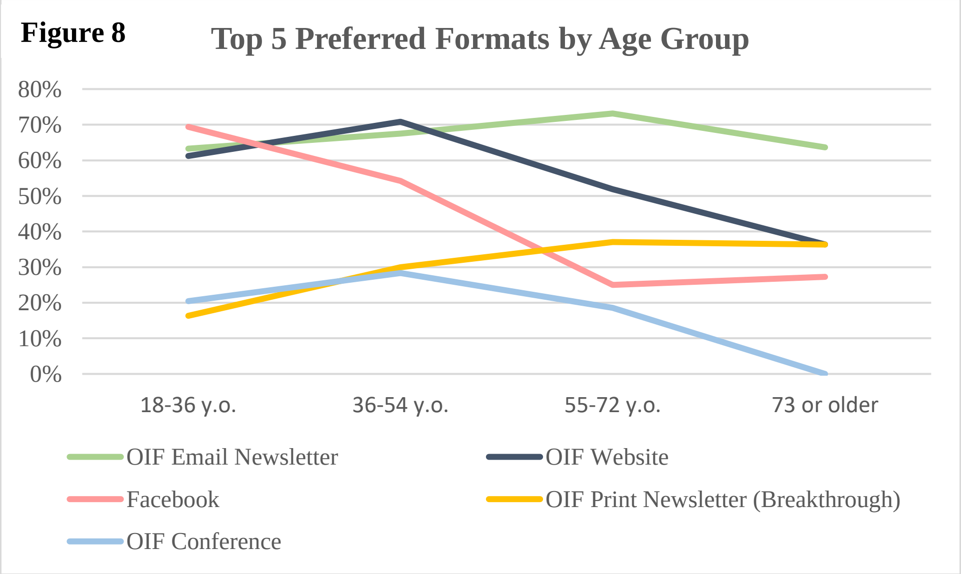


Figure 8 reveals positive slopes for 2 of the top 5 preferred formats (OIF Email Newsletter and OIF Print Newsletter Breakthrough) for information as age increases. See Figure 8 for complete distribution of preferred format rankings by age group.

Figure 8



Discussion

The results of the OIF Program Services Survey demonstrate a cost-efficient approach to implementing evidence-based patient education materials and programs. The number of total responses (385), survey completion rate (77%), and number of qualitative write-in responses indicate a highly engaged and motivated constituent population. The reach and representation of the survey, as shown through the range of international and geographically dispersed participants, aligns with trends of many other OI-focused surveys, such as the OI-Adult Natural History Initiative (OI-ANHI) study (Tosi et al., 2015).

One of the positive outcomes of this study was the identification of “new” or prominent subpopulations within the OI community. The needs of the population affected by mild OI was already known by the OI Foundation, as it is well-established that OI is an extremely heterogeneous disorder; however, the overwhelmingly large response requesting more information tailored for less severe types of OI was clear throughout the survey results. Another emerging subpopulation noted in the survey analysis is the parent population of grown children living with OI. The survey results document several comments from parents who felt that conference sessions did not apply to them because their children are now grown. In the past, the OI Foundation successfully organized “peer-to-peer discussion roundtables” for smaller subpopulations, such as grandparents or siblings of individuals living with OI. This could be a solution to address the needs of emerging subpopulations. The recognition of various subpopulations regarding the type of OI, age, and geographic location reinforces the idea of regional conferences and special topic workshops. The geographic diversity of respondents and comments about cost and transportation related challenges validate the need to implement more webinars and virtual activities.

The responses about preferred formats of accessing information about OI and the OIF were not expected. When designing the survey, members of the study team hypothesized that Facebook or another social media would rank as the most preferred format to receive information. The OIF email newsletter was ranked by 69% of survey respondents as their most preferred format, followed by the OIF website (59%). These results have significant implications on the communication strategies of the OIF.

Strengths

A major strength of this study was the response rate over a period of three weeks. The ability to access quick feedback is valuable for many purposes. The survey helped create awareness around newer initiatives, such as podcasts and the OI Educate Program. This study was successful in collecting data points on a wide range of topics, programs and publications, while providing a baseline structure and tool for future assessments.

Limitations

This survey was not designed with validated measures; therefore, it may not be reliable or replicable with another set of survey respondents within the OI population. There were limited published examples of needs assessment surveys in disease-specific and rare disease populations. Of the literature available on related topics, there were sporadic reports not published in academic journals or developed with IRB approval. Two members of the study team, including the Principal Investigator, are employed by the OI Foundation. The study team was careful to reduce the potential risk of bias.

An often-reoccurring theme in research is that women typically far outnumber men as respondents to surveys (Tosi et al., 2015). Though not surprising, more outreach to male community members is needed to increase their current 15% response rate to closer towards equally representative (50%). The overall response and completion rate was high; however, many survey respondents skipped questions, resulting in 23% incomplete survey responses.

Conclusion

The OIF Program Services Survey helped identify new topics and categories to implement in programs and publications. During the period of data collection and analysis, several inquiries from other rare disease organizations and international counterparts to the OIF requested the survey template. One of the goals of this project is to provide an effective survey instrument to share with other patient advocacy groups. Once another round of feedback from OIF and BBDC leadership is implemented and the manuscript is submitted, the template will be shared with other groups. The OIF would also like to implement an annual or biennial Program Services Survey as a shared international resource and recruit participants from more sources.

Setting priorities for programs and publications can be challenging for small patient advocacy groups with limited resources. Non-profits focused on rare disorders must plan and invest strategically in their program services to maximize the impact for all constituent communities. Results from the OIF Program Services Survey provided valuable recommendations to improve services and ensure that activities are well-aligned with the missions of the OI Foundation and the Brittle Bone Disorders Consortium. The survey results had an immediate effect on the design of upcoming communications and programs. Studies have shown that personal preferences on formats and timing of information may affect an individual's ability to retain and process information and their ability to engage with their care team, which could ultimately influence health outcomes (Jessup et al., 2016). Considerations must be made for the large variety of preferences in information formats, as shown by the survey results. It is critical that publications and programs are not difficult for patients to access, as it could exacerbate disparities for populations in the most need of information and support. A critical component to future success and impact of the OI Foundation is the involvement of volunteer leaders in the research and medical community (Hart et al., 2014). The OIF Program Services Survey confirmed the critical need for increased collaboration, effective communication, and forceful advocacy amongst all stakeholders (Hart et al., 2014).

As the science of health education and patient engagement evolves, data on health information consumers benefits the patient population in a variety of contexts. With the use of needs assessments, more effective methods of communication are identified for physicians and patient advocacy groups, and the constituent community and patient population are able to voice their most critical needs and concerns. The results of this survey reiterate the findings from a study published in the Journal of Pediatric Nursing, which emphasized the importance of “understanding social contexts, including families, peers, everyday activities, and relationships” in patient education and the development of informational materials (Driessnack, 2009). The OI Foundation is dedicated to building powerful partnerships between patients, scientists, and physicians and empowering a well-informed and “research ready” community.

Informational needs assessments and health literacy tools assist in understanding the dynamic needs of a patient population and larger constituent community. The OI Foundation serves as an advocate to expand the understanding of the causes and consequences of OI, and acts as a conduit to accelerate research for improved treatments and eventually a cure. As OI-related educational programs and services continue to reach new and larger populations, the OI Foundation will continue to build on the history of nearly five decades of mission-driven work and partnerships.

"The reward for work well done is the opportunity to do more." – Jonas Salk, MD

References

Anand, A., Tullis, E., Stephenson, A., & Abhyankar, P. (2014). Development and evaluation of an educational website for adults with cystic fibrosis. *Journal of Cystic Fibrosis*, *13*(3), 306-310.

Badiu, C., Bonomi, M., Borshchevsky, I., Cools, M., Craen, M., ... & Juul, A. (2017). Developing and evaluating rare disease educational materials co-created by expert clinicians and patients: the paradigm of congenital hypogonadotropic hypogonadism. *Orphanet journal of rare diseases*, *12*(1), 57. Retrieved from: https://link.springer.com/article/10.1186/s13023-017-0608-2.

Budych, K., Helms, T. M., & Schultz, C. (2012). How do patients with rare diseases experience the medical encounter? Exploring role behavior and its impact on patient–physician interaction. *Health Policy*, *105*(2), 154-164.

Cella, D., Yount, S., Rothrock, N., Gershon, R., Cook, K., Reeve, B., ... & Rose, M. (2007). The Patient-Reported Outcomes Measurement Information System (PROMIS): progress of an NIH Roadmap cooperative group during its first two years. *Medical care*, *45*(5 Suppl 1), S3.

Centers for Disease Control and Prevention (CDC). (2016). Health Literacy: Understanding Literacy and Numeracy. Retrieved from: https://www.cdc.gov/healthliteracy/learn/UnderstandingLiteracy.html

Cline, R. J., & Haynes, K. M. (2001). Consumer health information seeking on the Internet: the state of the art. *Health education research*, *16*(6), 671-692.

Chan, C. V., & Kaufman, D. R. (2011). A framework for characterizing eHealth literacy demands and barriers. *Journal of medical Internet research*, *13*(4).

Children's National. (2017, August 14). Giving voice to adult osteogenesis imperfecta patients. *Innovation District.* Retrieved from: https://innovationdistrict.childrensnational.org/giving-voice-adult-osteogenesis-imperfecta-patients/.

Childs, O. (2017, July 24). Health literacy 'elephant in the room' in patient engagement. *PharmaPhorum.* Retrieved from: https://pharmaphorum.com/views-and-analysis/health-literacy-elephant-room-patient-engagement/.

Driessnack, M. (2009). Growing up at the intersection of the genomic era and the information age. *Journal of pediatric nursing*, *24*(3), 189-193.

Egbert, N., & Nanna, K. M. (2009). Health literacy: Challenges and strategies. *The Online Journal of Issues in Nursing*, *14*(3). Retrieved from: http://www.nursingworld.org/MainMenuCategories/ANAMarketplace/ANAPeriodicals/OJIN/TableofContents/Vol142009/No3Sept09/Health-Literacy-Challenges.html.

Faster Cures. (2017, June). From Aspiration to Application: 5 Years of Patient Centricity. Retrieved from http://www.fastercures.org/assets/Uploads/Patients-Count-June-2017.pdf.

Feldman-Stewart, D., Brennenstuhl, S., & Brundage, M. D. (2007). A purpose-based evaluation of information for patients: an approach to measuring effectiveness. *Patient education and counseling*, *65*(3), 311-319. Retrieved from: https://www.ncbi.nlm.nih.gov/pubmed/17014979.

Food & Drug Administration (FDA). (2017). FDA awards six grants for natural history studies in rare diseases. *FDA Press Releases.* Retrieved from: https://www.fda.gov/NewsEvents/Newsroom/PressAnnouncements/ucm579375.htm.

Fox, S. (2011). The Social Life of Health Information. Pew Internet and American Life. Retrieved from: http://www.pewresearch.org/fact-tank/2014/01/15/the-social-life-of-health-information/.

Google.com. (2018). *Features-Google Analytics*. (online www.oif.org). Accessed March 5, 2018.

Hamilton, J. G., Hutson, S. P., Frohnmayer, A. E., Han, P. K., Peters, J. A., Carr, A. G., & Alter, B. P. (2015). Genetic information-seeking behaviors and knowledge among family members and patients with Inherited Bone Marrow Failure Syndromes. *Journal of Genetic Counseling*, *24*(5), 760-770.

Hart, T., Huber, M. The Osteogenesis Imperfecta Community and Scientific Research: A Valuable Partnership (23-27). In Shapiro JR. (Ed.) (2014). Osteogenesis Imperfecta: A Translational Approach to Brittle Bone Disease 1st Edition. New York, NY: Elsevier Academic Press.

Herder, M. (2017). What Is the Purpose of the Orphan Drug Act?. *PLoS medicine*, *14*(1), e1002191. Retrieved from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC5207521/.

Heijmans, M., Waverijn, G., Rademakers, J., van der Vaart, R., & Rijken, M. (2015). Functional, communicative and critical health literacy of chronic disease patients and their importance for self-management. *Patient Education and Counseling*, *98*(1), 41-48.

Jessup, M., Douglas, T., Priddis, L., Branch-Smith, C., & Shields, L. (2016). Parental experience of information and education processes following diagnosis of their infant with cystic fibrosis via newborn screening. *Journal of pediatric nursing*, *31*(3), e233-e241. Retrieved from: http://www.sciencedirect.com/science/article/pii/S0882596315003668.

Koohy, H., & Koohy, B. (2014). A lesson from the ice bucket challenge: using social networks to publicize science. *Frontiers in genetics*, *5*, 430.

Litzkendorf, S., Babac, A., Rosenfeldt, D., Schauer, F., Hartz, T., Lührs, V., ... & Frank, M. (2016). Information needs of people with rare diseases-what information do patients and their relatives require? *Journal of Rare Disorders: Diagnosis & Therapy*, *2*(2).

McLellan, F. (1998). “Like hunger, like thirst”: patients, journals, and the internet. *The Lancet*, *352*, S39-S43.

Mohamed, N. E., Herrera, P. C., Hudson, S., Revenson, T. A., Lee, C. T., Quale, D. Z., ... & Diefenbach, M. A. (2014). Muscle invasive bladder cancer: examining survivor burden and unmet needs. *The Journal of Urology*, *191*(1), 48-53. Retrieved from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4286331/.

Nakamura, C., Bromberg, M., Bhargava, S., Wicks, P., & Zeng-Treitler, Q. (2012). Mining online social network data for biomedical research: a comparison of clinicians’ and patients’ perceptions about amyotrophic lateral sclerosis treatments. *Journal of Medical Internet Research*, *14*(3).

National Library of Medicine. (2000). *Current Bibliographies in Medicine: Health Literacy.* Retrieved from: https://www.nlm.nih.gov/archive/20061214/pubs/cbm/hliteracy.html

Nielsen-Bohlman, L., Panzer, A. M., & Kindig, D. A. (2004). Health literacy: a prescription to end confusion. Committee on Health Literacy, Board on Neuroscience and Behavioral Health, Institute of Medicine. *Washington, DC: The National Academies Press.*

Nutbeam, D. (2009). Defining and measuring health literacy: what can we learn from literacy studies? *International Journal of Public Health, 54:303.* Retrieved from: https://link.springer.com/article/10.1007/s00038-009-0050-x.

Office of Disease Prevention and Health Promotion. (2010). National Action Plan to Improve Health Literacy [Internet]. Retrieved from: http://health.gov/communication/HLActionPlan/

OI Foundation. (n.d.). OIF Needs Assessment Survey. OI Foundation Archives, Gaithersburg, MD.

OI Foundation. (2017, November). Adults Who are Affected by OI. Retrieved from: http://www.oif.org/OIF_Adults.

Parker, R. M., Baker, D. W., Williams, M. V., & Nurss, J. R. (1995). The test of functional health literacy in adults. *Journal of general internal medicine*, *10*(10), 537-541.

Scotten, M. (2015). Parental health literacy and its impact on patient care. *Primary Care: Clinics in Office Practice*, *42*(1), 1-16. Retrieved from: http://www.primarycare.theclinics.com/article/S0095-4543(14)00105-5/abstract.

Siegfried, T. (2010). Odds Are, It's Wrong. *Science News: Magazine of the Society for Science & the Public*. Retrieved from: https://www.sciencenews.org/article/odds-are-its-wrong.

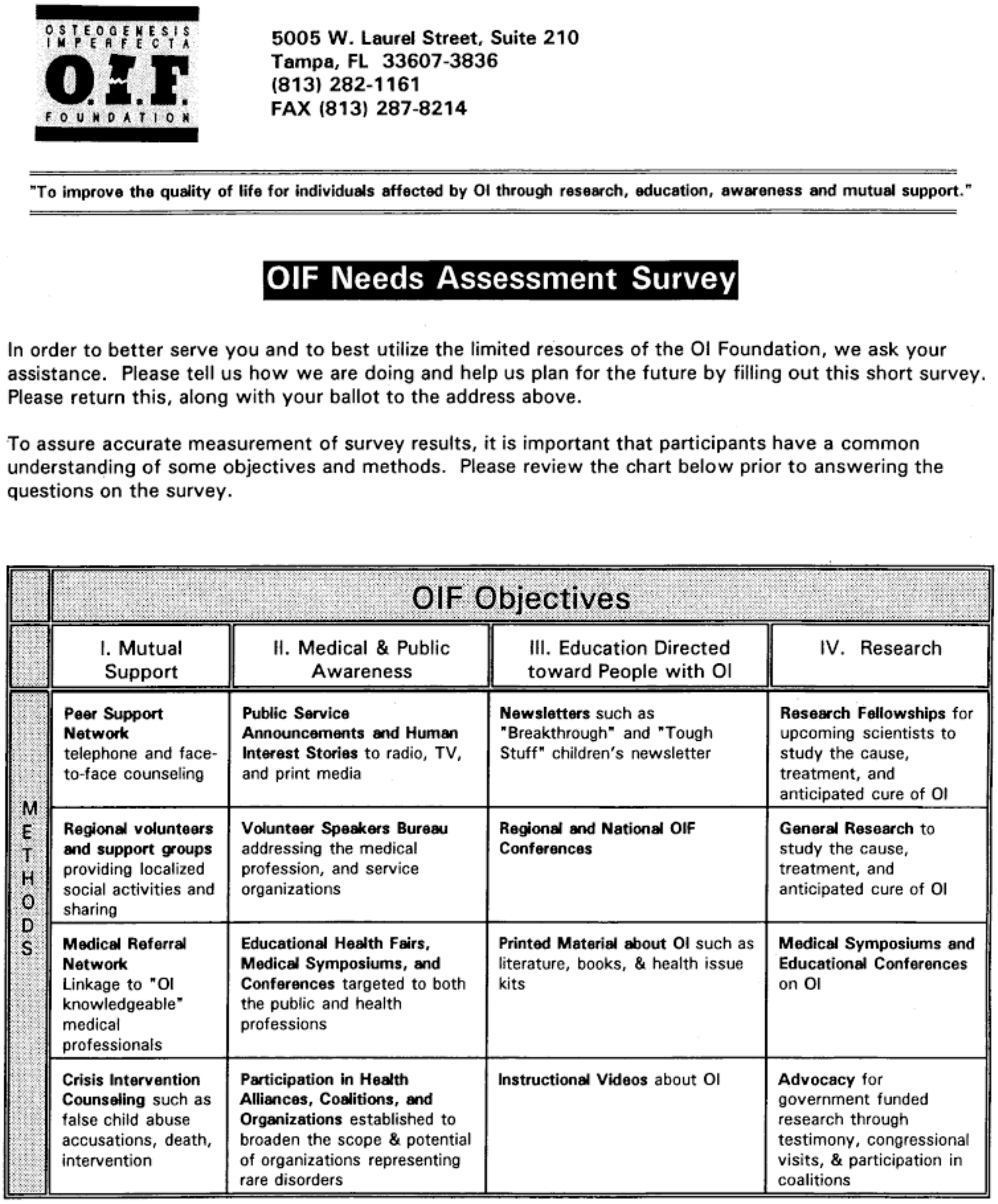
Tosi, L. L., Oetgen, M. E., Floor, M. K., Huber, M. B., Kennelly, A. M., McCarter, R. J., ... & McKiernan, F. E. (2015). Initial report of the osteogenesis imperfecta adult natural history initiative. *Orphanet journal of rare diseases*, *10*(1), 146. Retrieved from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4650321/pdf/13023_2015_Article_362.pdf.

Williams, M. V., Davis, T., Parker, R. M., & Weiss, B. D. (2002). The role of health literacy in patient-physician communication. *Family Medicine- Kansas City-*, *34*(5), 383-389.

Appendices

Appendix A – OIF Needs Assessment Survey

(OI Foundation, n.d.)



OSTEOGENESIS IMPERFECTA
O.I.F.
FOUNDATION

5005 W. Laurel Street, Suite 210
Tampa, FL 33607-3836
(813) 282-1161
FAX (813) 287-8214

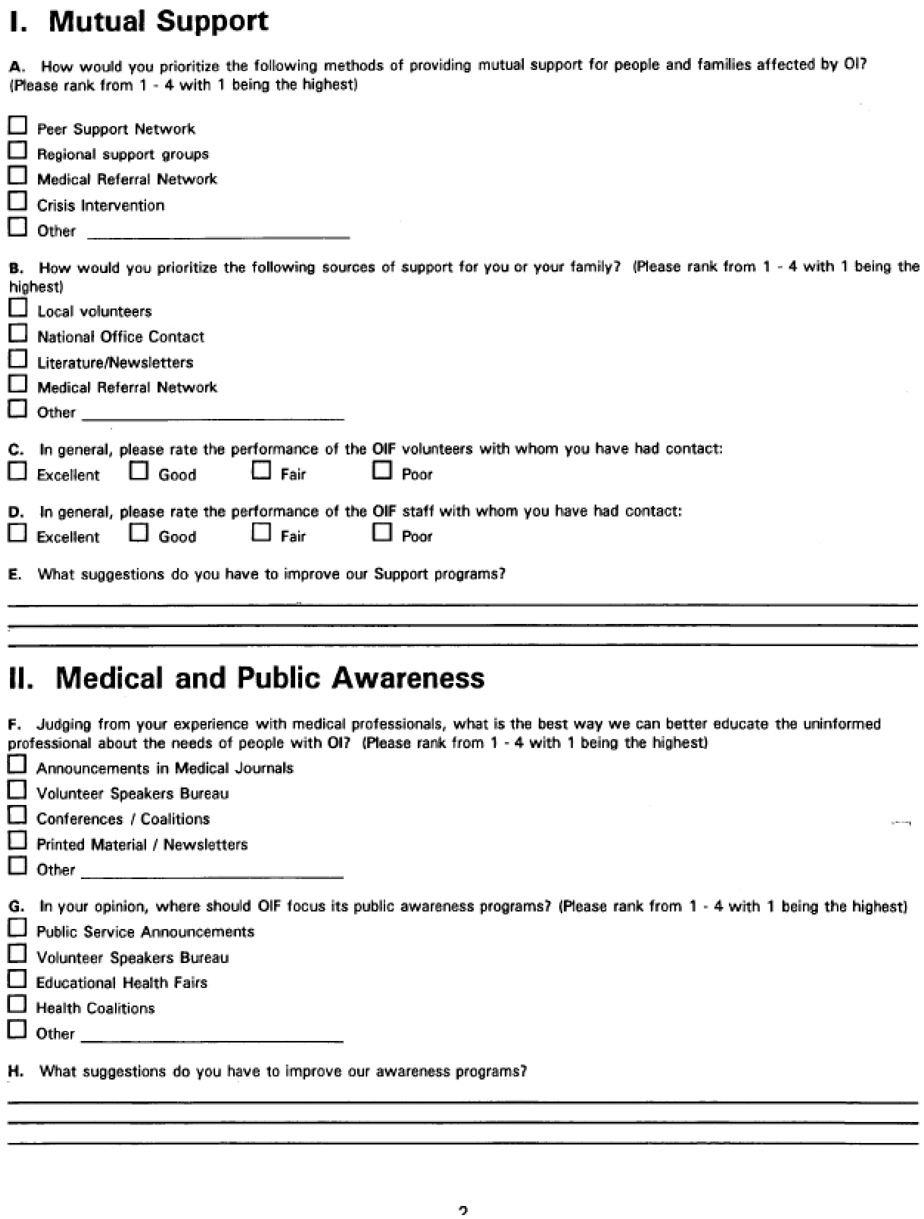
"To improve the quality of life for individuals affected by OI through research, education, awareness and mutual support."

OIF Needs Assessment Survey

In order to better serve you and to best utilize the limited resources of the OI Foundation, we ask your assistance. Please tell us how we are doing and help us plan for the future by filling out this short survey. Please return this, along with your ballot to the address above.

To assure accurate measurement of survey results, it is important that participants have a common understanding of some objectives and methods. Please review the chart below prior to answering the questions on the survey.

	OIF Objectives			
	I. Mutual Support	II. Medical & Public Awareness	III. Education Directed toward People with OI	IV. Research
METHODS	**Peer Support Network** telephone and face-to-face counseling	**Public Service Announcements and Human Interest Stories** to radio, TV, and print media	**Newsletters** such as "Breakthrough" and "Tough Stuff" children's newsletter	**Research Fellowships** for upcoming scientists to study the cause, treatment, and anticipated cure of OI
	Regional volunteers and support groups providing localized social activities and sharing	**Volunteer Speakers Bureau** addressing the medical profession, and service organizations	**Regional and National OIF Conferences**	**General Research** to study the cause, treatment, and anticipated cure of OI
	Medical Referral Network Linkage to "OI knowledgeable" medical professionals	**Educational Health Fairs, Medical Symposiums, and Conferences** targeted to both the public and health professions	**Printed Material about OI** such as literature, books, & health issue kits	**Medical Symposiums and Educational Conferences** on OI
	Crisis Intervention Counseling such as false child abuse accusations, death, intervention	**Participation in Health Alliances, Coalitions, and Organizations** established to broaden the scope & potential of organizations representing rare disorders	**Instructional Videos** about OI	**Advocacy** for government funded research through testimony, congressional visits, & participation in coalitions



I. Mutual Support

A. How would you prioritize the following methods of providing mutual support for people and families affected by OI? (Please rank from 1 - 4 with 1 being the highest)

☐ Peer Support Network
☐ Regional support groups
☐ Medical Referral Network
☐ Crisis Intervention
☐ Other ____________________

B. How would you prioritize the following sources of support for you or your family? (Please rank from 1 - 4 with 1 being the highest)

☐ Local volunteers
☐ National Office Contact
☐ Literature/Newsletters
☐ Medical Referral Network
☐ Other ____________________

C. In general, please rate the performance of the OIF volunteers with whom you have had contact:

☐ Excellent ☐ Good ☐ Fair ☐ Poor

D. In general, please rate the performance of the OIF staff with whom you have had contact:

☐ Excellent ☐ Good ☐ Fair ☐ Poor

E. What suggestions do you have to improve our Support programs?

____
____
____

II. Medical and Public Awareness

F. Judging from your experience with medical professionals, what is the best way we can better educate the uninformed professional about the needs of people with OI? (Please rank from 1 - 4 with 1 being the highest)

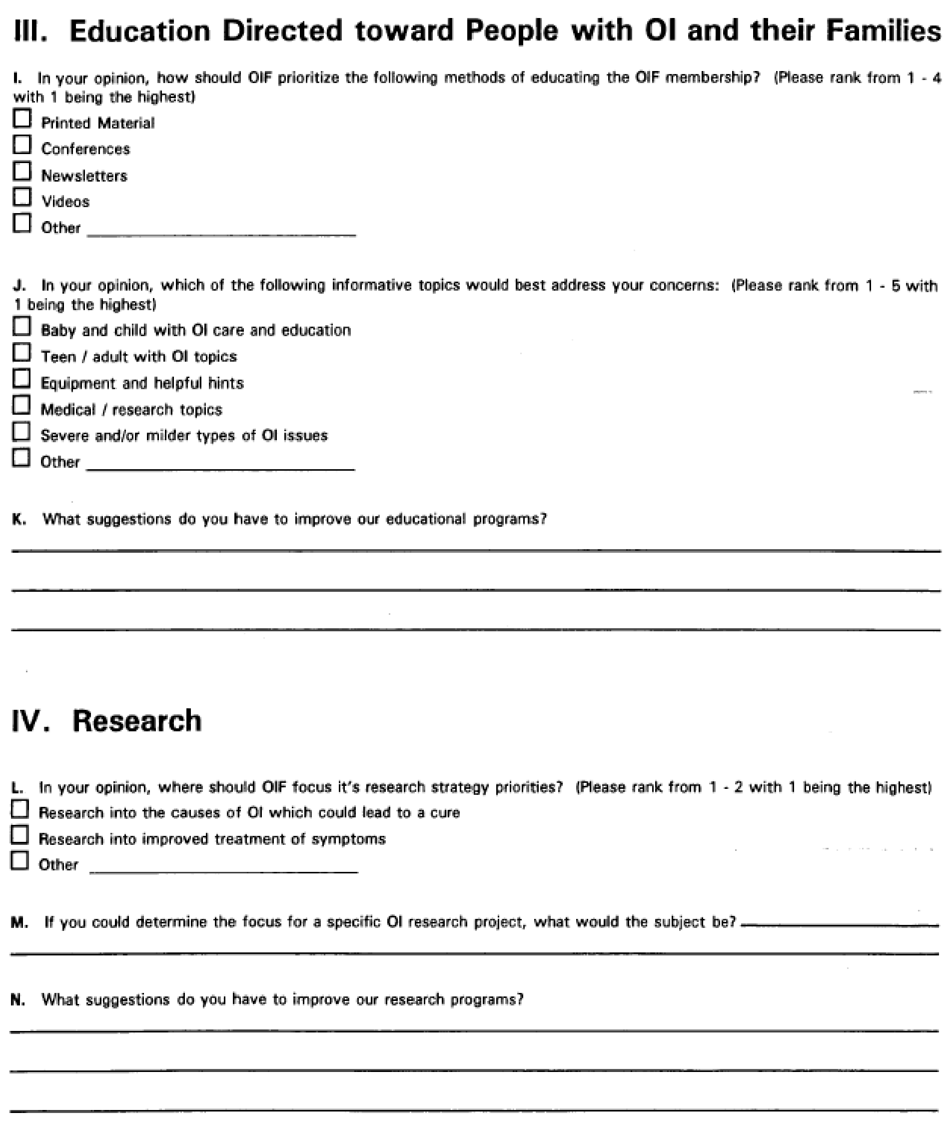
☐ Announcements in Medical Journals
☐ Volunteer Speakers Bureau
☐ Conferences / Coalitions
☐ Printed Material / Newsletters
☐ Other ____________________

G. In your opinion, where should OIF focus its public awareness programs? (Please rank from 1 - 4 with 1 being the highest)

☐ Public Service Announcements
☐ Volunteer Speakers Bureau
☐ Educational Health Fairs
☐ Health Coalitions
☐ Other ____________________

H. What suggestions do you have to improve our awareness programs?

____
____
____



III. Education Directed toward People with OI and their Families

I. In your opinion, how should OIF prioritize the following methods of educating the OIF membership? (Please rank from 1 - 4 with 1 being the highest)

☐ Printed Material
☐ Conferences
☐ Newsletters
☐ Videos
☐ Other ______________________________

J. In your opinion, which of the following informative topics would best address your concerns: (Please rank from 1 - 5 with 1 being the highest)

☐ Baby and child with OI care and education
☐ Teen / adult with OI topics
☐ Equipment and helpful hints
☐ Medical / research topics
☐ Severe and/or milder types of OI issues
☐ Other ______________________________

K. What suggestions do you have to improve our educational programs?

____

____

____

IV. Research

L. In your opinion, where should OIF focus it's research strategy priorities? (Please rank from 1 - 2 with 1 being the highest)

☐ Research into the causes of OI which could lead to a cure
☐ Research into improved treatment of symptoms
☐ Other ______________________________

M. If you could determine the focus for a specific OI research project, what would the subject be? ____________________

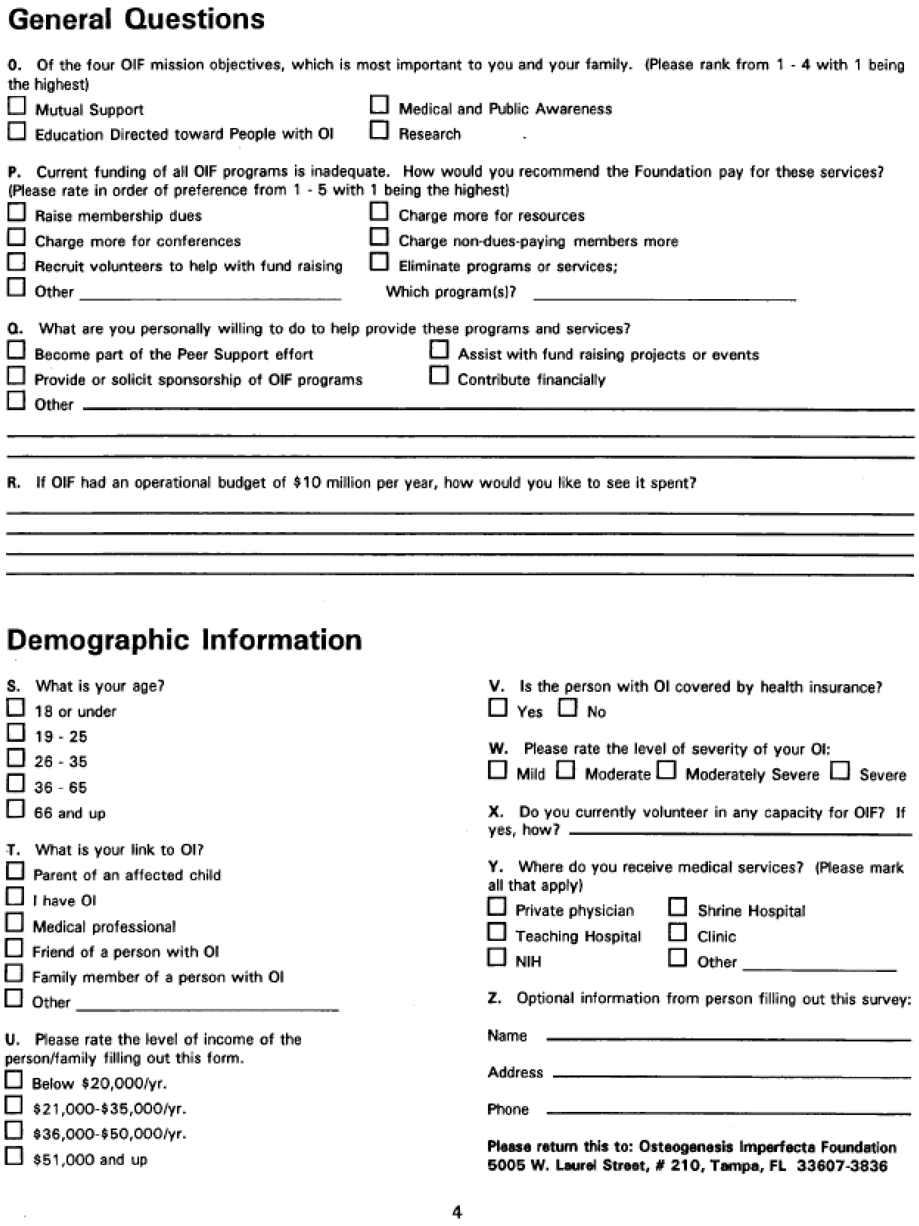
____

N. What suggestions do you have to improve our research programs?

____

____

____



General Questions

O. Of the four OIF mission objectives, which is most important to you and your family. (Please rank from 1 - 4 with 1 being the highest)

☐ Mutual Support
☐ Education Directed toward People with OI
☐ Medical and Public Awareness
☐ Research

P. Current funding of all OIF programs is inadequate. How would you recommend the Foundation pay for these services? (Please rate in order of preference from 1 - 5 with 1 being the highest)

☐ Raise membership dues
☐ Charge more for conferences
☐ Recruit volunteers to help with fund raising
☐ Other ____
☐ Charge more for resources
☐ Charge non-dues-paying members more
☐ Eliminate programs or services;
Which program(s)? ____

Q. What are you personally willing to do to help provide these programs and services?

☐ Become part of the Peer Support effort
☐ Provide or solicit sponsorship of OIF programs
☐ Other ____
☐ Assist with fund raising projects or events
☐ Contribute financially

R. If OIF had an operational budget of $10 million per year, how would you like to see it spent?

Demographic Information

S. What is your age?

☐ 18 or under
☐ 19 - 25
☐ 26 - 35
☐ 36 - 65
☐ 66 and up

T. What is your link to OI?

☐ Parent of an affected child
☐ I have OI
☐ Medical professional
☐ Friend of a person with OI
☐ Family member of a person with OI
☐ Other ____

U. Please rate the level of income of the person/family filling out this form.

☐ Below $20,000/yr.
☐ $21,000-$35,000/yr.
☐ $36,000-$50,000/yr.
☐ $51,000 and up

V. Is the person with OI covered by health insurance?

☐ Yes ☐ No

W. Please rate the level of severity of your OI:

☐ Mild ☐ Moderate ☐ Moderately Severe ☐ Severe

X. Do you currently volunteer in any capacity for OIF? If yes, how? ____

Y. Where do you receive medical services? (Please mark all that apply)

☐ Private physician
☐ Teaching Hospital
☐ NIH
☐ Shrine Hospital
☐ Clinic
☐ Other ____

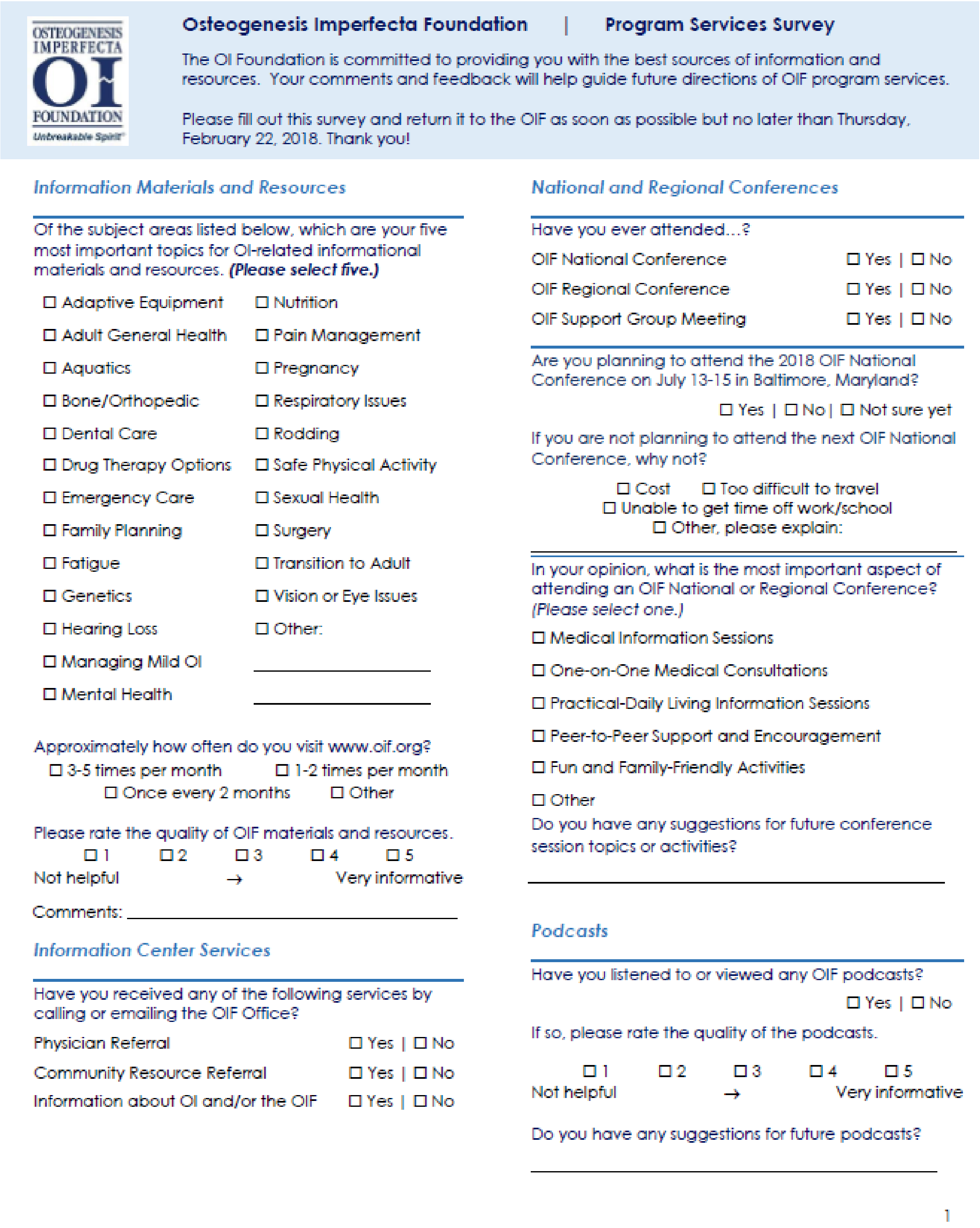
Z. Optional information from person filling out this survey:

Name ____
Address ____
Phone ____

Please return this to: Osteogenesis Imperfecta Foundation
5005 W. Laurel Street, # 210, Tampa, FL 33607-3836

4

Appendix B - Survey Instrument



Osteogenesis Imperfecta Foundation | Program Services Survey

The OI Foundation is committed to providing you with the best sources of information and resources. Your comments and feedback will help guide future directions of OIF program services.

Please fill out this survey and return it to the OIF as soon as possible but no later than Thursday, February 22, 2018. Thank you!

Information Materials and Resources

Of the subject areas listed below, which are your five most important topics for OI-related informational materials and resources. ***(Please select five.)***

☐ Adaptive Equipment
☐ Adult General Health
☐ Aquatics
☐ Bone/Orthopedic
☐ Dental Care
☐ Drug Therapy Options
☐ Emergency Care
☐ Family Planning
☐ Fatigue
☐ Genetics
☐ Hearing Loss
☐ Managing Mild OI
☐ Mental Health
☐ Nutrition
☐ Pain Management
☐ Pregnancy
☐ Respiratory Issues
☐ Rodding
☐ Safe Physical Activity
☐ Sexual Health
☐ Surgery
☐ Transition to Adult
☐ Vision or Eye Issues
☐ Other:
____
____

Approximately how often do you visit www.oif.org?
☐ 3-5 times per month ☐ 1-2 times per month
☐ Once every 2 months ☐ Other

Please rate the quality of OIF materials and resources.
☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5
Not helpful → Very informative

Comments: ________________________________

Information Center Services

Have you received any of the following services by calling or emailing the OIF Office?

Physician Referral	☐ Yes | ☐ No
Community Resource Referral	☐ Yes | ☐ No
Information about OI and/or the OIF	☐ Yes | ☐ No

National and Regional Conferences

Have you ever attended...?

OIF National Conference	☐ Yes | ☐ No
OIF Regional Conference	☐ Yes | ☐ No
OIF Support Group Meeting	☐ Yes | ☐ No

Are you planning to attend the 2018 OIF National Conference on July 13-15 in Baltimore, Maryland?
☐ Yes | ☐ No | ☐ Not sure yet

If you are not planning to attend the next OIF National Conference, why not?
☐ Cost ☐ Too difficult to travel
☐ Unable to get time off work/school
☐ Other, please explain:
____

In your opinion, what is the most important aspect of attending an OIF National or Regional Conference? *(Please select one.)*

☐ Medical Information Sessions
☐ One-on-One Medical Consultations
☐ Practical-Daily Living Information Sessions
☐ Peer-to-Peer Support and Encouragement
☐ Fun and Family-Friendly Activities
☐ Other

Do you have any suggestions for future conference session topics or activities?
____

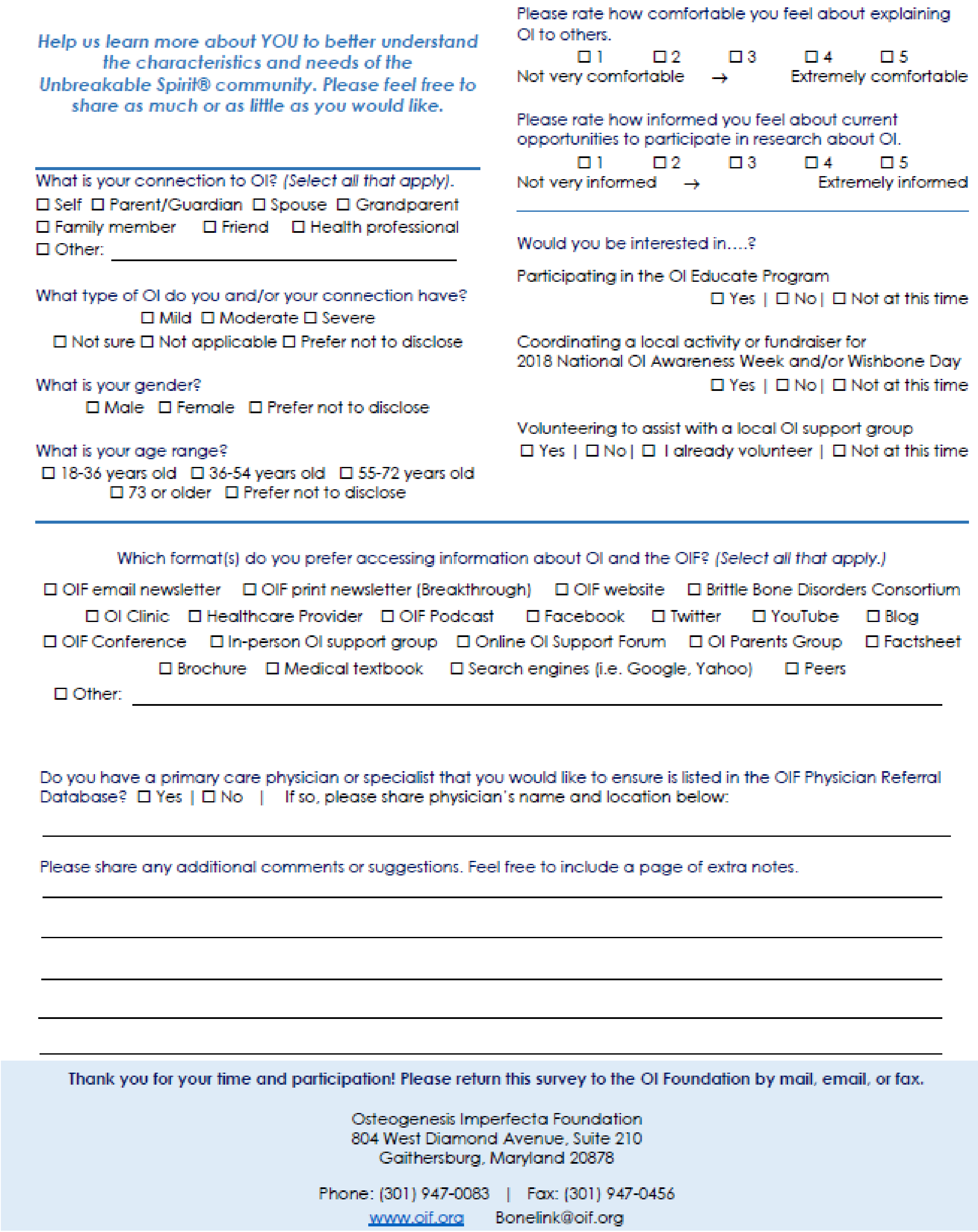
Podcasts

Have you listened to or viewed any OIF podcasts?
☐ Yes | ☐ No

If so, please rate the quality of the podcasts.
☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5
Not helpful → Very informative

Do you have any suggestions for future podcasts?
____

1



Help us learn more about YOU to better understand the characteristics and needs of the Unbreakable Spirit® community. Please feel free to share as much or as little as you would like.

What is your connection to OI? *(Select all that apply).*
☐ Self ☐ Parent/Guardian ☐ Spouse ☐ Grandparent
☐ Family member ☐ Friend ☐ Health professional
☐ Other:

What type of OI do you and/or your connection have?
☐ Mild ☐ Moderate ☐ Severe
☐ Not sure ☐ Not applicable ☐ Prefer not to disclose

What is your gender?
☐ Male ☐ Female ☐ Prefer not to disclose

What is your age range?
☐ 18-36 years old ☐ 36-54 years old ☐ 55-72 years old
☐ 73 or older ☐ Prefer not to disclose

Please rate how comfortable you feel about explaining OI to others.
☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5
Not very comfortable → Extremely comfortable

Please rate how informed you feel about current opportunities to participate in research about OI.
☐ 1 ☐ 2 ☐ 3 ☐ 4 ☐ 5
Not very informed → Extremely informed

Would you be interested in….?

Participating in the OI Educate Program
☐ Yes | ☐ No | ☐ Not at this time

Coordinating a local activity or fundraiser for 2018 National OI Awareness Week and/or Wishbone Day
☐ Yes | ☐ No | ☐ Not at this time

Volunteering to assist with a local OI support group
☐ Yes | ☐ No | ☐ I already volunteer | ☐ Not at this time

Which format(s) do you prefer accessing information about OI and the OIF? *(Select all that apply.)*
☐ OIF email newsletter ☐ OIF print newsletter (Breakthrough) ☐ OIF website ☐ Brittle Bone Disorders Consortium
☐ OI Clinic ☐ Healthcare Provider ☐ OIF Podcast ☐ Facebook ☐ Twitter ☐ YouTube ☐ Blog
☐ OIF Conference ☐ In-person OI support group ☐ Online OI Support Forum ☐ OI Parents Group ☐ Factsheet
☐ Brochure ☐ Medical textbook ☐ Search engines (i.e. Google, Yahoo) ☐ Peers
☐ Other:

Do you have a primary care physician or specialist that you would like to ensure is listed in the OIF Physician Referral Database? ☐ Yes | ☐ No | If so, please share physician's name and location below:

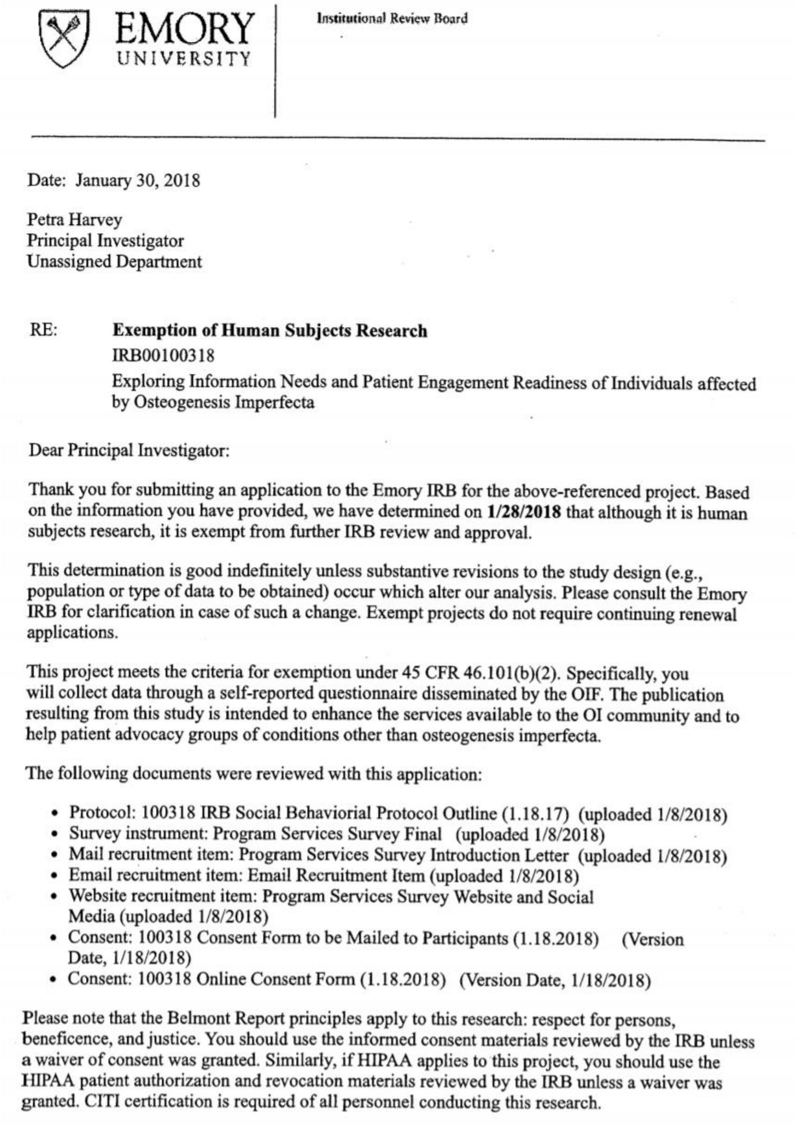
Please share any additional comments or suggestions. Feel free to include a page of extra notes.

Thank you for your time and participation! Please return this survey to the OI Foundation by mail, email, or fax.

Osteogenesis Imperfecta Foundation
804 West Diamond Avenue, Suite 210
Gaithersburg, Maryland 20878

Phone: (301) 947-0083 | Fax: (301) 947-0456
www.oif.org Bonelink@oif.org

Appendix C - IRB Approval letter



EMORY
UNIVERSITY

Institutional Review Board

Date: January 30, 2018

Petra Harvey
Principal Investigator
Unassigned Department

RE: **Exemption of Human Subjects Research**
IRB00100318
Exploring Information Needs and Patient Engagement Readiness of Individuals affected by Osteogenesis Imperfecta

Dear Principal Investigator:

Thank you for submitting an application to the Emory IRB for the above-referenced project. Based on the information you have provided, we have determined on **1/28/2018** that although it is human subjects research, it is exempt from further IRB review and approval.

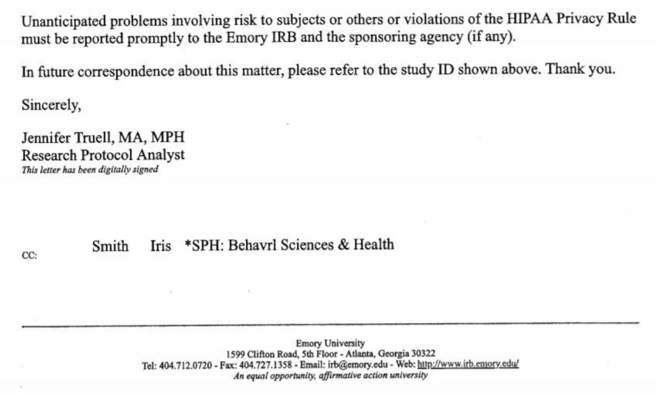
This determination is good indefinitely unless substantive revisions to the study design (e.g., population or type of data to be obtained) occur which alter our analysis. Please consult the Emory IRB for clarification in case of such a change. Exempt projects do not require continuing renewal applications.

This project meets the criteria for exemption under 45 CFR 46.101(b)(2). Specifically, you will collect data through a self-reported questionnaire disseminated by the OIF. The publication resulting from this study is intended to enhance the services available to the OI community and to help patient advocacy groups of conditions other than osteogenesis imperfecta.

The following documents were reviewed with this application:

- Protocol: 100318 IRB Social Behaviorial Protocol Outline (1.18.17) (uploaded 1/8/2018)
- Survey instrument: Program Services Survey Final (uploaded 1/8/2018)
- Mail recruitment item: Program Services Survey Introduction Letter (uploaded 1/8/2018)
- Email recruitment item: Email Recruitment Item (uploaded 1/8/2018)
- Website recruitment item: Program Services Survey Website and Social Media (uploaded 1/8/2018)
- Consent: 100318 Consent Form to be Mailed to Participants (1.18.2018) (Version Date, 1/18/2018)
- Consent: 100318 Online Consent Form (1.18.2018) (Version Date, 1/18/2018)

Please note that the Belmont Report principles apply to this research: respect for persons, beneficence, and justice. You should use the informed consent materials reviewed by the IRB unless a waiver of consent was granted. Similarly, if HIPAA applies to this project, you should use the HIPAA patient authorization and revocation materials reviewed by the IRB unless a waiver was granted. CITI certification is required of all personnel conducting this research.



Unanticipated problems involving risk to subjects or others or violations of the HIPAA Privacy Rule must be reported promptly to the Emory IRB and the sponsoring agency (if any).

In future correspondence about this matter, please refer to the study ID shown above. Thank you.

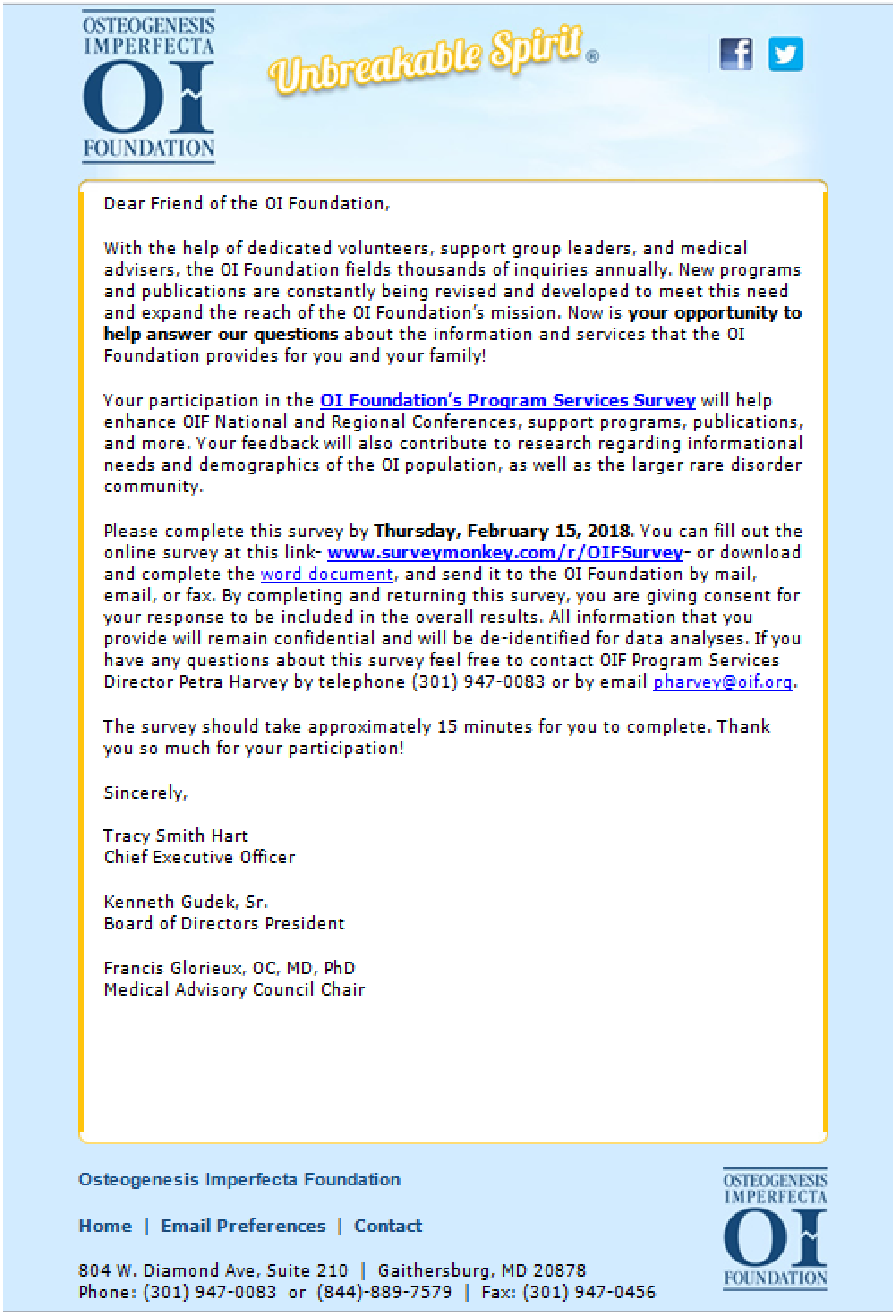
Sincerely,

Jennifer Truell, MA, MPH
Research Protocol Analyst
This letter has been digitally signed

cc: Smith Iris *SPH: Behavrl Sciences & Health

Emory University
1599 Clifton Road, 5th Floor - Atlanta, Georgia 30322
Tel: 404.712.0720 - Fax: 404.727.1358 - Email: irb@emory.edu - Web: http://www.irb.emory.edu/
An equal opportunity, affirmative action university

Appendix D - Recruitment Email



Dear Friend of the OI Foundation,

With the help of dedicated volunteers, support group leaders, and medical advisers, the OI Foundation fields thousands of inquiries annually. New programs and publications are constantly being revised and developed to meet this need and expand the reach of the OI Foundation's mission. Now is **your opportunity to help answer our questions** about the information and services that the OI Foundation provides for you and your family!

Your participation in the **OI Foundation's Program Services Survey** will help enhance OIF National and Regional Conferences, support programs, publications, and more. Your feedback will also contribute to research regarding informational needs and demographics of the OI population, as well as the larger rare disorder community.

Please complete this survey by **Thursday, February 15, 2018**. You can fill out the online survey at this link- **www.surveymonkey.com/r/OIFSurvey**- or download and complete the word document, and send it to the OI Foundation by mail, email, or fax. By completing and returning this survey, you are giving consent for your response to be included in the overall results. All information that you provide will remain confidential and will be de-identified for data analyses. If you have any questions about this survey feel free to contact OIF Program Services Director Petra Harvey by telephone (301) 947-0083 or by email pharvey@oif.org.

The survey should take approximately 15 minutes for you to complete. Thank you so much for your participation!

Sincerely,

Tracy Smith Hart
Chief Executive Officer

Kenneth Gudek, Sr.
Board of Directors President

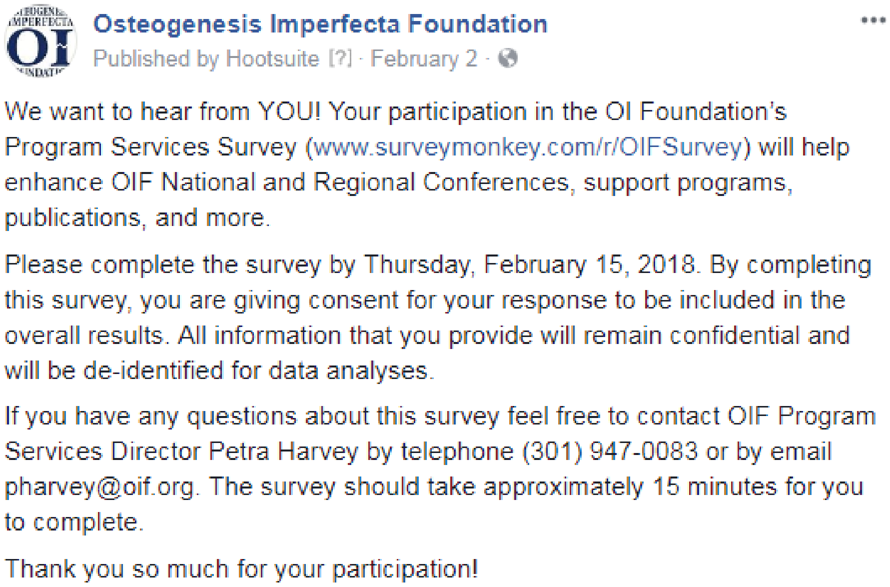
Francis Glorieux, OC, MD, PhD
Medical Advisory Council Chair

Osteogenesis Imperfecta Foundation

Home | Email Preferences | Contact

804 W. Diamond Ave, Suite 210 | Gaithersburg, MD 20878
Phone: (301) 947-0083 or (844)-889-7579 | Fax: (301) 947-0456

Appendix E - Social Media Recruitment Posts



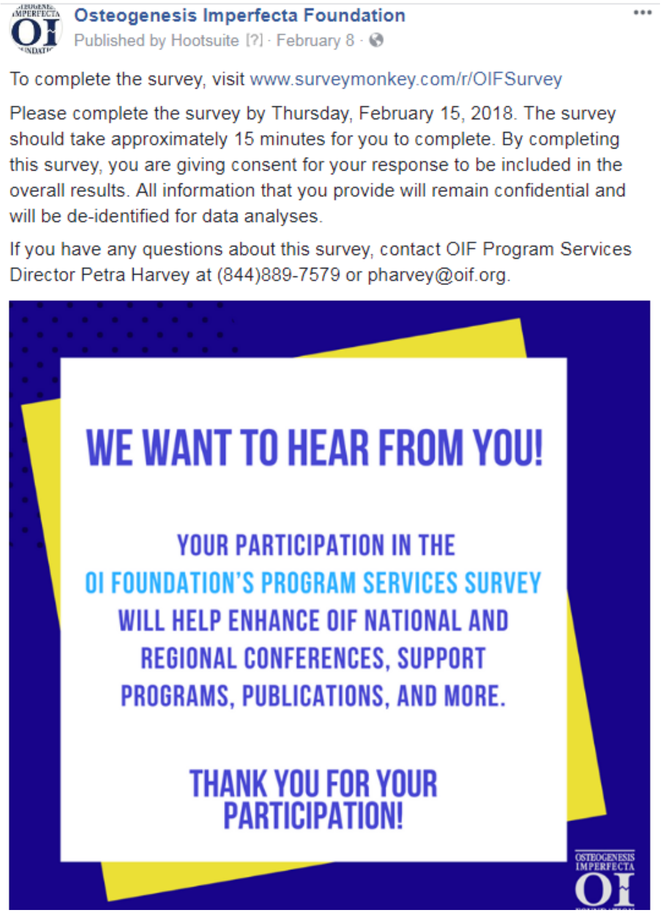
Osteogenesis Imperfecta Foundation
Published by Hootsuite [?] · February 2 ·

We want to hear from YOU! Your participation in the OI Foundation's Program Services Survey (www.surveymonkey.com/r/OIFSurvey) will help enhance OIF National and Regional Conferences, support programs, publications, and more.

Please complete the survey by Thursday, February 15, 2018. By completing this survey, you are giving consent for your response to be included in the overall results. All information that you provide will remain confidential and will be de-identified for data analyses.

If you have any questions about this survey feel free to contact OIF Program Services Director Petra Harvey by telephone (301) 947-0083 or by email pharvey@oif.org. The survey should take approximately 15 minutes for you to complete.

Thank you so much for your participation!



Osteogenesis Imperfecta Foundation
Published by Hootsuite [?] · February 8 ·

To complete the survey, visit www.surveymonkey.com/r/OIFSurvey

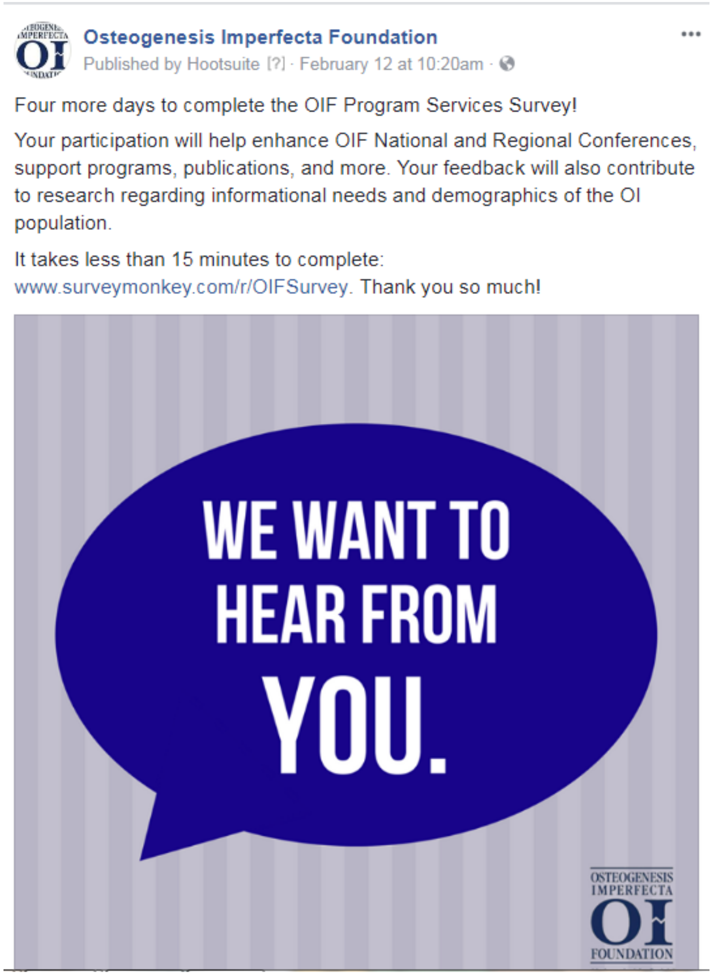
Please complete the survey by Thursday, February 15, 2018. The survey should take approximately 15 minutes for you to complete. By completing this survey, you are giving consent for your response to be included in the overall results. All information that you provide will remain confidential and will be de-identified for data analyses.

If you have any questions about this survey, contact OIF Program Services Director Petra Harvey at (844)889-7579 or pharvey@oif.org.

WE WANT TO HEAR FROM YOU!

YOUR PARTICIPATION IN THE OI FOUNDATION'S PROGRAM SERVICES SURVEY WILL HELP ENHANCE OIF NATIONAL AND REGIONAL CONFERENCES, SUPPORT PROGRAMS, PUBLICATIONS, AND MORE.

THANK YOU FOR YOUR PARTICIPATION!



Osteogenesis Imperfecta Foundation
Published by Hootsuite [?] · February 12 at 10:20am ·

Four more days to complete the OIF Program Services Survey!

Your participation will help enhance OIF National and Regional Conferences, support programs, publications, and more. Your feedback will also contribute to research regarding informational needs and demographics of the OI population.

It takes less than 15 minutes to complete: www.surveymonkey.com/r/OIFSurvey. Thank you so much!

OSTEOGENESIS IMPERFECTA
OI
FOUNDATION
Unbreakable Spirit®

WE WANT TO HEAR FROM YOU!

WWW.SURVEYMONKEY.COM/R/OIFSURVEY

**COMPLETE THE
OIF PROGRAM SERVICES
SURVEY TODAY**