

Title

The day-to-day experiences of caring for children with Osteogenesis Imperfecta: A qualitative descriptive study

Abstract

Aims and objectives: This study aimed to explore the day-to-day experiences of family caregivers who are caring for children with Osteogenesis Imperfecta (OI).

Background: OI is a rare genetic condition known to cause bone fragility. Family caregivers of children with OI play an important role in helping these children live well at home.

Design: A qualitative descriptive design was used.

Methods: A qualitative descriptive study was conducted in accordance with the COREQ guidelines. Adult family caregivers (n=18) of children with OI were recruited from a university-affiliated, paediatric orthopaedic hospital in Montreal, Canada. Individual interviews were conducted, transcribed verbatim, and inductively thematically analysed.

Results: OI family caregiving entailed: (a) managing regular day-to-day caregiving activities, including morning routines, evening routines, and the facilitation of their child's mobilization; (b) coping with periods that made the caregiving routine more challenging, such as fractures, surgeries, and pain; and (c) devising long-term strategies to support day-to-day care, such as managing the

1 environment, accessing medical and school resources, and coordinating and
2 accessing respite care.

3 **Conclusions:** The day-to-day routine of caring for a child with OI may be
4 disrupted by challenging periods and improved by long-term strategies developed
5 to ease day-to-day care. These strategies suggest future directions for clinicians
6 and policy makers to improve health services and caregiver well-being.

7 **Relevance to clinical practice:** Clinical, policy, and research endeavours need to
8 incorporate new interventions to support the needs of family caregivers. These
9 recommendations may be relevant to other clinicians and policymakers working
10 with families living with rare and chronic physical conditions.

11 **Keywords:** care needs, carers, child nursing, chronic illness, community care,
12 parenting, paediatrics, orthopaedics

Introduction

Osteogenesis Imperfecta (OI) is a rare genetic condition affecting 1 in 10 000 births that is predominantly caused by an alteration in the genes responsible for collagen production (Marini et al., 2017; Trejo & Rauch, 2016). Osteogenesis Imperfecta is characterized by increased bone fragility leading to frequent fractures, but this chronic condition can also cause teeth and soft tissue abnormalities, discoloured sclera, hearing loss, shortened stature, and skeletal deformities (Marini et al., 2017; Trejo & Rauch, 2016). Family caregivers, such as parents, play a critical role in helping children with unique needs thrive at home and in the community (McCann, Bull, & Winzenberg, 2012). Understanding the day-to-day experiences of caregivers of children with rare conditions, including caregivers of children with OI, is imperative to inform the development of policies and services tailored to their needs (McCann et al., 2012). Therefore, the aimed to explore the day-to-day experiences of family caregivers (e.g. parents and legal guardians) caring for children with OI.

Background

There are several OI classifications with a range of phenotypes and severity. OI Types I – IV are the original classifications of OI, although other types have been discovered since (Trejo & Rauch, 2016). OI Type I is the most common form of the condition and has the mildest disease severity, with fewer fractures expected over the life course. OI Type II is lethal in the neonatal period. OI Type III is the most severe form of OI that is compatible with a longer lifespan. OI Type IV is of moderate disease severity. OI Types V, VI, and VII are newer classifications that present with different tissue and bone mineralization phenotypes, and have mild to moderate severity (Marini et al., 2017; Trejo & Rauch, 2016). At present, there is no cure for OI for this life-long condition (Chougui et al., 2020). Bisphosphonate therapy is used to improve bone mass

density (Trejo & Rauch, 2016). Surgery, physical therapy, and occupational therapy are other common treatment modalities (Dogba et al., 2013; Marini et al., 2017).

When a child has OI, a parent usually adopts the role of primary caregiver to assist with their child's everyday needs (Vanz, Felix, da Rocha, & Schwartz, 2015). Challenges unique to OI caregiving include coping with the constant risk of fractures, balancing the need for over-protectionism, adjusting to social isolation, adapting the home and other environments, and dealing with potential suspicions of physical child abuse (Arabaci et al., 2015; Bernehall & Brodin, 2002; Dogba, Rauch, Tre, Glorieux, & Bedos, 2014). Parents of children with OI have reported lower scores in the environmental quality of life domain (e.g., environmental safety and access to social supports) compared to the general parental population (Szczepaniak-Kubat, Kurnatowska, Jakubowska-Pietkiewicz, & Chlebna-Sokol, 2012; Vanz et al., 2015). However, the OI caregiving experience may also generate positive experiences for families, such as the development of an optimistic mindset and being inspired by the child's resilience (Dogba et al., 2013). Yet, for many families, a child's OI diagnosis appears to be a life-altering event requiring various and ongoing parental adjustments (Arabaci et al., 2015; Dogba et al., 2013).

While several studies on the OI family experience have included some analysis of the day-to-day lives of OI caregivers (Bernehall & Brodin, 2002; Dogba et al., 2013; Dogba et al., 2014; Hill, Baird, & Walters, 2014; Santos, Pires, Soares, & Barros, 2017), none of these studies focused primarily on the minutiae and routines of daily care. Thus, this study sought to better understand the daily experiences of family caregivers (e.g. parents and legal guardians) of children living with OI.

Methods

Design

The study methodology was qualitative description (Sandelowski, 2000). This study was part of a larger study where the primary aim was to explore the views of OI family caregivers on using Internet-based technologies to support their caregiving needs (Castro, Chougui, Bilodeau, & Tsimicalis, 2019).

Sample and Recruitment

This study was conducted at a university-affiliated, bilingual (English and French), paediatric orthopaedic hospital in Montreal, Canada. A purposive sampling strategy was used to recruit family caregivers of children who were being treated for any type of OI at the study site. (Sandelowski, 2000) suggests a research team decides on sample size based on judgement, applied research methods, and experience in the research area. Based on our principal investigator's past research with OI families, our sample size of 15-18 caregivers was deemed adequate for achieving "a new and richly textured understanding of experience" of OI caregiving (Sandelowski, 1995, p. 183).

Inclusion criteria included all family caregivers of patients treated for OI at the study site, who were the child's primary caregiver, and who spoke either English or French. A non-authoritative member of the team, CB, approached clinicians with a scheduled appointment with the OI patients to mediate a study introduction. If caregivers expressed an interest in learning more about the study, a member of the research team was notified to further describe the study and if appropriate, to obtain informed consent, and to schedule an interview based on the caregiver's convenience.

Ethical Considerations

Ethical approval was received in the spring of 2017 from McGill University's Institutional Review Board (IRB) prior to study set-up and recruitment. The IRB conforms with the Canadian Tri-Council Policy Statement on the Ethical Conduct for Research Involving Humans. All participants provided written informed consent to participate in the study, with the understanding their personal information would not be identifiable in any research publications. All participant data were anonymized and kept confidential.

Data Collection

A demographic data survey was completed by each participant. Individual semi-structured interviews were subsequently conducted by a member of the research team, in either English or French depending on the caregiver's preference. The interviews were conducted by either author AC or KC, at the study site or by telephone or videoconferencing. Both researchers conducting interviews had past experience in qualitative research, and one of them has OI. The principal investigator has past experience conducting both quantitative and qualitative interview-based studies. Key questions relevant to day-to-day care included in the interview guide of the larger study are listed in Table 1. The interview guide was reviewed by the OI clinical team prior to data collection. Interviews were audiotaped and transcribed with field notes recorded during and after the interviews. A reflective journal was kept by author AC throughout the duration of the study.

Analysis

The demographic surveys were analyzed using descriptive statistics. The transcribed data and field notes were uploaded into Microsoft Excel, where the data

were first inductively open-coded by three of the authors (AC, KC, and JM), and then further analyzed primarily by AC with inter-coder checks by KC using (Braun & Clarke, 2006)'s six steps for thematic analysis. Interviews conducted in French were first open coded into English by authors KC and JM who are bilingual. In frequent meetings with the principal investigator (AT), the research team discussed the ongoing development of the codebook by AC and established the final themes. These codebooks, meeting notes, and ongoing reflections were recorded in an anonymized, reflective journal.

Rigour

Qualitative rigour was established using (Lincoln & Guba, 1985)'s four criteria of credibility, dependability, confirmability, and transferability. Credibility was enhanced by the use of reflexive journaling, the audio-taping of interviews, and the frequent research meetings for peer debriefing (Polit & Beck, 2012). The research dependability was enhanced by careful documentation of our research team's discussions and decisions in the reflexive journal (available for audit upon request) (Polit & Beck, 2012). The research confirmability was supported by documenting the decision trail in the reflexive journal, hosting regular team meetings and debriefs, and keeping an audit trail (Polit & Beck, 2012). The research transferability was supported by the development of analytical materials such as the codebook, inter-coder checks, and the use of thick descriptions and quotes (Polit & Beck, 2012). The Consolidated Criteria for Reporting Qualitative Research (COREQ) guided the study including the data collection, analysis, and reporting of the research (Supplementary File 1) (Tong, Sainsbury, & Craig, 2007).

Results

Demographic Data

Nineteen caregivers were approached in-person, and all verbally agreed to participate. One international caregiver was lost to follow-up following hospital discharge. Eighteen adult caregivers from 14 families were individually interviewed. Caregivers were divided into mother and father figures, allowing for the inclusion of either biological parents or legal guardians. Some of their children with OI required no mobilizing assistance, while others used a wheelchair full-time. Some parents had children with few fractures and surgeries; however, the child of one mother had experienced over 200 fractures. Interviews ranged from 15 minutes to 1.5 hours in length. The 18 interviews resulted in 13.05 hours of audio-recordings and over 8000 spreadsheet lines of coded transcript. Further demographic data are listed in Table 2.

Caregiving Themes

The following themes relating to the day-to-day experiences of OI caregiving were inductively identified: managing regular day-to-day caregiving activities, coping with challenging periods, and devising long-term strategies caregivers developed to support day-to-day care. By the end of the analysis of our final 18 interviews, no new major themes were identified, suggesting that thematic saturation was adequately achieved for this sample of caregivers.

Managing regular day-to-day caregiving activities

OI caregivers were asked to share what a regular OI caregiving day looked like. Key components of these day-to-day activities included following morning and evening

1 routines to assist with their child's activities of daily living (ADLs), and facilitating
2 mobilization.

3 ***Morning and evening routines.*** Mornings were particularly busy for caregivers,
4 when many ADLs tended to occur all at once, and all tasks needed to be completed
5 before school and paid work began. Parents emphasized the importance of having a
6 routine to manage the mornings. As one mother explained:

7 The stress of the morning routine, it's big . . . planning and taking care of, make
8 sure I don't forget anything if we go out, the wheelchair, the emergency kit, I
9 mean it's always ready but...
10 (Mother, Family 5, Type III)
11

12 Mornings consisted of waking everyone up, making lunches, helping children get
13 dressed, and using the bathroom. Some children needed assistance in dressing with their
14 leg braces or corsets. Children needing the most assistance with ADLs were younger
15 children and those with more severe types of OI. Chronic pain management was also a
16 part of the morning routine for at least two families. Most families chose to drive their
17 children to school, although one child used an adapted bus. One mother said her son
18 would walk to school if it was nice outside, but if his backpack was heavy or the
19 sidewalk was icy, she would drive him to school, instead:

20 Um, his backpack with all of his things and his laptop is now super heavy. We
21 live in [a northern region], so the majority of the time is icy, slippery. If I didn't
22 drive him, he'd walk. So there's no ... like, we are only about two blocks away
23 from the school, so if it is really nice outside sometimes he walks home from
24 school with friends, but I would go and pick up his backpack.
25 (Mother, Family 13, Type III)
26

27 Another family explained that wheelchair ramps were often too icy or snowy during
28 winter to be useful:

29 So, we go down the ramp, and we put him in the truck, you lift him up, we go to
30 the school ... and where we live, it's the Arctic, so it's winter 10 months out of
31 the year. So, the amount of snow we get is ridiculous. They don't always have
32 the wheelchair ramp, you know, snow-cleared.
33 (Mother A, Family 8, Type VI)

Other caregivers did not use the adapted school bus as well because the bus arrived too early during their busy mornings. So, sometimes, services meant to be accessible were not truly accessible for these families' daily routines.

Evenings were described as being more relaxed than mornings:

And then once we get back home, it's okay, 'cause I don't feel the pressure of not being late to work and not being late to school. I have the pressure of like putting the beds, putting the kids to bed earlier, and [OI child] is getting older, so it's not as stressful. He's a bit more independent, more mature, so he needs a little bit less attention,
(Mother, Family 5, Type III)

In the evenings, caregivers made dinner, had their children complete chores, played games with their children, talked about school, and engaged in their personal low-impact family activities – such as listening to audiobooks as a family:

We like to listen to audiobooks, so we just YouTube a lot of audiobooks ... It's our go-to bedtime routine, we put a good book on and we kind of fall asleep listening to a book.
(Father, Family 4, Type I)

During the evenings, caregivers also took their children to specialized and frequent physical therapy and occupational therapy appointments. School-aged children were expected to use some of the evening time for schoolwork. A few parents specifically commended their children's academic and social successes:

I'm proud of my kids. [Child with OI] does super well at school and has a nice personality.
(Mother, Family 5, Type III)

He, like, he's really joyful. And even when he has difficulties, um when he has a fracture and being like . . . he's not a dramatic boy, he doesn't complain, he doesn't ask for much. He just wants me by his side or holding more during those days, but - he still has joy.
(Mother, Family 9, Type III)

Some children engaged in adapted sports, various artistic classes, and other low-impact activities, such as swimming and bocchia ball. Older children with fewer accessibility challenges independently played outside with friends in the evenings.

Facilitating mobilization. Mobilizing or transferring their children during these day-to-day activities was conducted carefully. Some school-aged children crawled or slid around on their buttocks, reserving walkers and wheelchairs for fatiguing times.

One child with a milder form of OI benefited from regular encouragement to independently move, particularly up stairs where she experienced greater difficulties.

Transfers presented with some of the greatest challenges for families. Many children required at least some assistance in transferring to and from the bed, car, wheelchair, and/or toilet:

Um. a regular day. Um ... because [son with OI] is not ... fully independent, and he's not very mobile, so . . . I guess I have to do his transfers for him. So that means a lot of lifting and positioning, either putting him in his small wheelchair, or putting him in his safe play area, all of that.
(Mother, Family 9, Type III)

We find...I think he [son with OI] finds having arm fractures more difficult, and I find him having leg fractures more difficult. So, a little bit different, just having to lift him a little bit more. To transfer, he's getting bigger, so he's getting heavier now. Like, in and out of the vehicle, especially with the leg fracture when it's new, those kind of things so ... that's a little bit different.
(Mother, Family 13, Type III)

One woman expressed “just moving him from point a to point b is most challenging”

(Mother B, Family 8, Type VI). While these caregivers typically lifted their school-aged child into the truck, this task was becoming more difficult as the child became heavier.

For families with children with more severe OI types, the stressful acts of transferring and mobilizing needed to be accomplished without causing further pain or fractures. In

contrast, families with Type I OI generally did not notice any significant differences in the day-to-day requirements of caring for their child living with OI.

1 *Coping with challenging periods*

2 OI caregivers were constantly aware of how their regular, daily routines for their
3 child with OI could be disrupted by fractures, surgeries, and pain. These challenging
4 periods increased the care management needed.

5 **Fractures.** The initial stress of bone breaks arose from the unexpectedness. The
6 child's first fracture(s) resulted in some caregiver panic, although caregivers learned to
7 adapt. One mother shared that her challenging day encompassed being called due to a
8 fracture at school:

9 The challenging time is when I'm called from the office that there's a fracture.
10 Sometimes she fractures in school, sometimes she fractures when she comes
11 back. And I will have to quickly rush home to go see the severity of the fracture,
12 whether it's just a crack, or it's a real fracture where we have to go to the
13 hospital. So basically, that's when it becomes challenging.
14 (Mother, Family 6, Type III)
15

16 She explained disliking her local hospitals in her respective country because the
17 clinicians lack knowledge of OI. She also felt stigmatized in hospitals.

18 With more severe forms of OI, a fracture may happen spontaneously, creating a
19 constant fear of fractures and feelings of stress upon fracture. A mother explained that
20 her fear of fractures halted her child's opportunity for play with other children:

21 Yes. That is the most challenging part of it. Allowing her to play with the other
22 kids . . . Because those ones are younger than her. They don't even know what
23 she has. They don't know that she has OI. They don't even know that she has
24 fracture. They don't even know what [a] fracture is. So you have to herd those
25 ones . . . stopping them from playing with her.
26 (Mother, Family 12, Type IV)
27

28 One caregiver explained how easily femur fractures could occur:

29 He was jumping on my bed. And, you know, like a normal four-year-old would
30 do. We were doing laundry, he was jumping on the bed, and with that, we heard
31 a *snap*. And he is *screaming*, leg bevelled out, and we're like "well, that's
32 strange." And then we went to the hospital, and they're like, "okay, it's a femur
33 fracture." And that happened about two other times after that. Then they're like,
34 "okay, we're going to test him [for OI]".
35 (Mother A, Family 8, Type VI)

Another caregiver shared that whenever her child fractured, she immediately assessed the fracture, administered appropriate medications, and adopted rest and relaxation for the remainder of the day. Other parents shared that in the case of severe fractures, they would go to the hospital and wait for treatment and recovery.

Surgeries. Surgeries presented unique difficulties for families. Hospital boredom was often a challenge for parents, especially if they had to fly to the study site for surgery and wait several days or weeks before returning home. A few caregivers discussed how challenging arranging surgical dates around their work and family schedules could be, as well as their need to sometimes accommodate hospital postponements. Care coordination for treatment created additional challenges for the three international families interviewed. Caregivers shared that accessing OI treatment in their own countries could be difficult. A few of them received pro-bono treatment supplies through the international OI community. For a mother whose daughter needed a few surgeries over several months, finding a Montreal sponsor was a significant care coordination challenge for her. She said she had to somehow find a sponsor to live with for several months, in a city far from her own, while also arranging care with her husband and their other children who would stay behind in her home country. She explained:

So I leave [the other children] with my husband when I came to the [Montreal OI treatment centre] in August . . . I think from the way it is, [the treatments] will end by December . . . You understand, I have my kids calling me, ‘Mommy, when are you coming back?’ . . . it’s a very long time.
(Mother, Family 12, Type IV)

The mother’s quote was similar to that of another international participant who explained that at times, he was unable to take time off from paid work to accommodate the long trips and surgical wait times. Even for families living in Canada, if they lived far from a hospital, the travel time and resources to access treatment restricted which

1 family member joined their child. As one participant, whose child always had to be
2 flown to the nearest hospital, explained:

3 Especially when you run into something... you know, we go to [major city
4 hospital] and they say, you know he's going to need surgery. They will not fly
5 you home if your appointment is within two weeks. So, you could be in the city
6 up to three, four weeks. (Mother A, Family 8, Type VI)

7
8 So, the time taken to access specialized surgical treatment was sometimes a cause of
9 separation among families.

10
11 ***Caregiving after a fracture or surgery.*** Caregiving became more difficult after a
12 fracture or a surgery, with the child's limb or limbs immobilized and unable to bear any
13 weight. Immobility required more assistance with ADLs, making transfers more
14 challenging. One mother explained that with a fractured extremity, her child became
15 non-weight-bearing:

16 Um, he's not able to propel his wheelchair on his own. He's not able to walk, so
17 that would mean more assistance for him when it comes to transfers, when he
18 wants to get from place to place, you know.
19 (Mother, Family 9, Type III)

20
21 Leg fractures were particularly problematic because the child became non-weight-
22 bearing on that limb, making transfers to and from places - such as the bathtub, toilet, or
23 car - heavy and complex for the caregiver.

24 When children fractured, time for self-care and outside excursions became more
25 difficult for caregivers, as well:

26 Especially when they're fractured, it's not as easy to get out. You don't want to
27 be transferring them when they're in pain, getting them into their car-seat. And
28 when you're the only person at home 'cause your husband's working, if you
29 need to go to the grocery store, then you have to take them with you. So
30 sometimes you're not necessarily getting yourself ready in the morning because
31 you're just trying to get them into a position where they can get out of the house
32 at some point or for an appointment.
33 (Mother, Family 13, Type III)

34

After surgery, the family's routine would change. For example, one mother explained how her child would sleep in her bedroom, receive more pain medication, and remain home for a few days post-surgery. A father noted that after his toddler's major surgeries, caregiving changed to reduce her activity:

Now, more recently, she was trying to get up. She got her surgery two weeks ago for OI. Specifically, what they did was they broke her tibia and her femur, and put in rodding for both of them. And then she was immobilized for ... well, her left leg's completely immobilized. And for her, that was different, because it changes everything for [toddler's name] because she used to be able to crawl everywhere where she wanted . . . So, the care kind of changed for her recently, because she . . . couldn't move as much.
(Father, Family 1, Type IV)

Fractures and surgeries created stress for caregivers during ADLs, as they were worried about any jostling during care work that could harm the child. As a result, caregivers tended to minimize activity and stay at home during these periods.

Pain. For some families of children with OI, life revolved around pain management. One mother explained that when her school-aged son was in pain, he would call for "Mommy!" a lot more frequently (Mother, Family 5, Type III). Another mother shared that her child was often in physical pain, particularly experiencing regular headaches and back pain:

[My daughter] has a lot of pain too . . . at least two or three times a day I have to assess her pain, with her, to see if I need to give medicine.
(Mother, Family 2, Type IV [Translated from French])

Two other parents also shared that their children experienced regular back pain in spite of their fractures being healed. One father explained, "I mean, ever since that first fracture, she's experiencing some pain...um...[in the same spot] like in her lower back – in her lower lumbar region. If she plays too much, she starts to get sore there" (Father, Family 4, Type I). Periods of increased pain created frequent disruptions to OI caregivers' daily routines.

Devising long-term strategies for supporting day-to-day care

A comment by one mother embodied the management of day-to-day care:

Dealing with OI is most likely all the time, how are we going to do things? You know? How are we going to surpass this limitation?

(Mother, Family 5, Type III)

Caregivers became experts in caring for their children's unique daily health and care needs. Caregivers developed strategies which would make future day-to-day caregiving work easier. They learned to manage the environment, access medical and school resources, and coordinate care and respite to support their caregiving over the long-term.

Managing the environment. Generally, caregivers tried to balance their desire to protect their child from harm, while also minimizing the limits placed on their child day-to-day. Managing care for caregivers often meant "managing the environment more than the kid":

We make sure that there is no chair or anything that could actually fall on her. the remote for the TV are never actually accessible if she's standing from the couch, because . . . it's heavy. It's an old remote. We do play stuff differently, without saying "no you can't" or "don't do that" or "don't touch that". It's really just managing the environment more than the kid . . . [The] environment is our responsibility.

(Mother, Family 1, Type IV)

Managing the environment included adapting their housing to be safer (for instance, by installing extra-cushioned carpeting), coordinating with schools to manage the care of their child safely, and searching for low-impact activities for their children. Managing the environment also entailed restricting their children to avoid certain physical activities. For instance, one father allowed his child to engage in most activities except for high impact ones, such as jumping on trampolines:

For the most part, it's the same as a regular child. Um...from wake up to routine to general activities...the only thing we watch out for is her playtime – no trampolines, that's off-limits.

(Father, Family 4, Type I)

1
2 With these environmental management strategies, future day-to-day care work was
3 made easier, because their homes and schools were more prepared for a child with
4 brittle bones to thrive in, and their children thrived in low-impact activities.

5 *Accessing medical and school supports.* Caregivers described spending a lot of
6 their caregiving time researching information and resources about OI, particularly for
7 accessing medical and school supports. Once specialized medical and school supports
8 were established, caregivers felt more at ease in their day-to-day care work.

9 *Accessing medical support.* For several caregivers, a diagnosis of OI did not
10 equate to adequate medical support and information for ongoing care. Healthcare
11 providers outside of the treatment centre – such as emergency staff and general
12 pediatricians at other hospitals – did not always know how to treat OI. Some parents
13 shared how other health professionals with limited OI experience would fracture their
14 child's bones in an effort to treat them. One mother believed insufficient information
15 was initially shared with her about her rights and available resources. She explained that
16 when her daughter was a pre-schooler, the two of them spent an entire year avoiding all
17 outings because they did not know how to mobilize her growing daughter beyond using
18 a baby stroller. They remained socially isolated and home-bound until learning that her
19 daughter had the right to access a properly fitted wheelchair. Caregivers shared that
20 once they connected with the OI experts from the study site, they subsequently felt
21 medically supported:

22 Well, uh, it was horrible, actually. They [healthcare providers in the family's
23 home state] couldn't even know what's going on, so they thought we were
24 abusing the child, and there was a lot of problems. But . . . when they [finally]
25 diagnosed it, everything cooled down. And they [his children] started coming
26 over here [to the treatment site], and getting easier, and a lot helpful over here
27 in Montreal.

28 (Father, Family 3, Type IV).
29

One family who moved to Montreal from their home country for work around the time of their child's diagnosis felt lucky to live near a specialized treatment centre:

It was a big move for us, but ... you know ... it has just been...amazing, being seen at the [study site / treatment site]. And, um, like having the best care for him. Even at that time we didn't even know yet why it was the best and... we didn't know yet. We sort of grew into that knowledge after ... after God provided us with a way to get here.
(Mother, Family 9, Type III)

Because OI is – OI is such a rare condition, it's...it's hard . . . it's mind-blowing for [the study site / treatment site] to specialize in OI, because it's rare. And that's not the case, especially in our country.
(Mother, Family 9, Type III)

Access to the greater OI community was an important resource for finding long-term medical supports. Several caregivers spent significant time seeking resources through their OI network. For instance, a few caregivers managed to receive regular OI treatment materials via the international OI community when these were not accessible in their own country. Once caregivers managed to access long-term OI medical supports, either with an established OI social network or – ideally – with a specialized OI treatment centre nearby, caregivers felt more supported in their day-to-day caregiving activities, because they had access to people knowledgeable about OI.

Accessing school supports. For some families, finding daycare centres and schools that were comfortable with a child with OI was a challenging task. Once they found an accommodating school, several families described making plans to go into the school every year to share with the new educators how to care for a child with OI. One mother shared that these new school caregivers had to be taught:

How do we immobilize a fracture, how do we give the medication, and how do we put him on the toilet seat . . . how do we take care of him?
(Mother, Family 5, Type III)

A father also noted that parents of children with OI have to educate school providers on their children's unique needs:

1 It's a little more stressful because as a parent you're always a little more worried
2 for her because, okay, if she does fall down. Ok, we're worried it's gonna be a
3 break. . . . You know, we have to inform the schools, we have to inform the
4 daycares, to be careful, that minor spills could be more – are a little more stressful
5 than, you know, [for] someone that doesn't have OI.

6 (Father, Family 4, Type I)

7
8 Most children were attending public schools. However, they still required access to a
9 school aide person who could help them in the washroom, carry books, push the
10 wheelchair, or write for them, as needed. A few caregivers were using more specialized
11 school services. One child moved to Montreal with her mother to attend a school for
12 children with special needs, while the child's father and siblings remained in their
13 hometown several hours away. Another family was choosing to homeschool their child
14 instead of sending him to a public school, because they wanted to ensure he was
15 mobilizing enough during the day. These caregivers made long-term choices – whether
16 it was to teach accommodating schools how to care for their child, or to homeschool
17 their child themselves – which would help them in their day-to-day care in the future.
18 By providing preventative education to school personnel early on, the personnel were
19 prepared for addressing the child's future care needs; by homeschooling their child to
20 ensure he was mobilizing enough daily, his physical strength would improve over time.

21 Still, other routine medical and school resource challenges included the
22 availability of services, language barriers, and Internet access. Even among financially
23 secure families, some families could not find appropriate daycare centres for their child
24 with more severe OI. One mother described the long waiting list for daycare for
25 children with special needs. Language barriers affected resource accessibility and access
26 to reliable Internet services. Some families noted that OI resources available in
27 languages outside of English (such as French) were limited. Families suggested that
28 access to the greater OI community through the Internet could also determine whether
29 families managed to find a specialized OI treatment centre and other OI resources.

1 ***Coordinating care and accessing respite.*** Caregivers described many care
2 coordination tasks related to their child's OI. These included the following tasks:
3 arranging and attending numerous medical appointments and treatments; remembering
4 to bring the wheelchair or walker whenever they went out; managing respite care; being
5 flexible with schedules in case of surgery or fractures; keeping track of medical records;
6 seeking out community and health resources; and filling out medical paperwork and
7 reimbursement forms. All of these activities required significant time and coordination
8 skills on the part of caregivers, which frustrated some of them. However, once these
9 coordination activities were accomplished, caregivers' day-to-day care work became
10 easier. For instance, by carefully organizing and keeping track of their child's medical
11 records, and by being flexible with their schedules, caregivers were able to quickly
12 adapt if medical emergencies arose.

13 Most caregivers expressed chronic challenges with accessing respite or short
14 break care, explaining that they did not trust others to care for their child with OI.
15 Several caregivers shared they were uncomfortable hiring a babysitter for their child
16 with OI, or even leaving their child with friends or family:

17 It's sort of an issue. You have to learn to trust, you have to make sure they
18 understand the needs, and don't over-do or not do at all.
19 (Mother, Family 1, Type IV).
20

21 My mother-in-law is yes, a very good help to us, but we don't normally have a
22 relaxed mind anywhere we are, outside home. So, you have to do anything you
23 are doing outside FAST. And come back.
24 (Mother, Family 12, Type IV)
25

26 Still, some families relied on grandparents and friends as caregivers and taught them to
27 care for their child, permitting some parenting breaks. Two mothers suggested that more
28 trusted short break care opportunities, such as kids' events or summer camps organized
29 by OI treatment centres, would be very helpful. If caregivers had easy access to people
30 they trusted to care for their child, day-to-day care was made easier because caregivers

1 had a reliable back-up caregiver. However, most caregivers did not have anyone they
2 fully trusted to provide this care.

3 **Discussion**

4 This study offered a comprehensive overview of the day-to-day experiences of
5 caring for a child with brittle bones, the challenging periods that can disrupt day-to-day
6 routines, and the long-term strategies parents developed to make day-to-day OI
7 caregiving easier. Understanding the unique daily experiences of caregivers of children
8 with rare and chronic conditions such as OI is important knowledge for helping
9 clinicians and policymakers to build better supports for these caregivers. Such supports
10 could improve caregivers' daily experiences of caregiving, and therefore improve their
11 child's care (Luijkx, Putten, & Vlaskamp, 2017; McCann et al., 2012).

12 The limited existing OI literature suggests that OI parents often have more tasks
13 to accomplish in one day than most parents do. Even on typical days without fractures
14 or surgery, a key difference is the child's independence in completing ADLs. For more
15 severe types of OI, in particular, the multiple physical transfers needed for toileting,
16 clothing, and transport increased the energy and amount of time required to accomplish
17 these ADLs (Dogba et al., 2014). Facilitating mobilization and transportation for those
18 living with rare and physical conditions is a common but challenging caregiving task
19 (Andrews, 2013). However, in addition to managing the task of mobilization, OI
20 caregivers of children with more severe types of OI also experience the unique and
21 constant fear of fracturing their child while completing transfers and other day-to-day
22 tasks (Dogba et al., 2014). Our study corroborates these reports of the experiences of
23 regular OI day-to-day caregiving, particularly the complex tasks of mobilization and
24 transfers.

1 For OI caregivers in our study, periods which presented particular challenges to
2 OI caregivers' day-to-day routines were periods with fractures, surgeries, and pain.
3 These time periods were also described as some of the most challenging aspects of OI
4 care in other studies (Dogba et al., 2013; Santos et al., 2017). However, while the
5 caregivers in our study acknowledged facing challenges and at times feeling
6 overwhelmed, most appeared to be successfully managing as parents and caregivers.
7 Caregivers explained that once they found the study site (a specialized OI treatment
8 centre), they felt more supported as caregivers. These results correspond with the results
9 of another study which was also conducted at our study site (Dogba et al., 2013).
10 However, these results contrast with other OI caregiving studies which described
11 caregivers feeling burned out, hopeless, and overwhelmed by the unique challenges of
12 caring for children with brittle bones (Arabaci et al., 2015; Bozkurt et al., 2014). These
13 latter studies may not have been conducted in settings with access to specialized OI
14 services. This contrast may speak to the psychosocial benefits that access to specialized
15 OI medical resources can have on OI families.

16 Managing the environment, accessing medical and school supports, coordinating
17 care, and finding trusted respite services are common care management strategies of
18 chronic caregivers (Pelentsov, Laws, & Esterman, 2015; Samia, O'Sullivan, Fallon,
19 Aboueissa, & Hepburn, 2018). Other researchers have noted that OI caregivers spend
20 significant amounts of time trying to gain reliable access to OI medical and school
21 supports, social supports, and respite care (Arabaci et al., 2015; Bernehall & Brodin,
22 2002; Bozkurt et al., 2014). These activities for facilitating day-to-day OI care over the
23 long-term were described by caregivers in our study, as well.

Strengths and Limitations

The methodological strengths of this study included providing rich descriptions of the study setting, participant demographics, and methods for transferability purposes. Our study benefited from having a research team member who is an expert in the subject matter, having lived with OI herself. The study results help address the paucity of literature available on OI caregiving, as well as on the supportive care needs of parents of children with a rare condition (Pelentsov et al., 2015). While corroborating much of the current OI caregiving literature, our study also adds to our new knowledge of OI caregiving by providing more detailed data on what day-to-day OI caregiving entails. Understanding the daily life of family caregivers is important because the differences required in care for caregivers of children with unique health needs can cause significant caregiver distress (Pelentsov et al., 2015). Understanding their unique needs can support the development of evidence-based health and social supports for these families (Collins et al., 2016; Luijkx et al., 2017).

This study has several limitations. The sample could have benefited from more socioeconomic caregiver diversity. Although thematic saturation was achieved with our sample, further studies should continue to explore the experiences of single parent families, those with lower socioeconomic status, the experiences of younger caregivers, and those who do not have access to sites specializing in OI management, as our sample only had a small representation of these families. Further research may seek to fully explore what an excellent or optimal day entails for caregivers of children with OI and use these study findings as preliminary work to better understand the diverse range of OI caregivers' day-to-day caregiving experiences.

Conclusion

The rich data collected from these interviews helped elucidate the day-to-day activities and challenges faced by OI caregivers. The results complement previous research in the caregiving and OI research fields, while also adding a deeper understanding of the day-to-day experiences of OI caregivers. This knowledge may help clinicians and healthcare systems offer improved services to these families. Findings provide further direction for future research and policy initiatives to help promote the health and well-being of OI caregivers.

Relevance to Clinical Practice

This study has implications for nurses and other paediatric clinicians. Finding information about OI, accessing treatment (especially for families who lived far away from the treatment centre), finding knowledgeable local healthcare providers, and coordinating available community and school supports, were all challenges faced by the caregivers in our study. Clinicians should compile and share the long-term strategies developed by expert OI caregivers with newly-diagnosed families with OI. Offering these strategies, may help the initial diagnosis be less traumatic, and give families practical information needed to care for their child at home in the community (Dogba et al., 2013). They should also work to develop clinical home-care tools to provide long-term supports for OI families. For instance, the study site has supported the development of a splint kit that helps caregivers splint their children's fractures at home. This kit is particularly valuable for families and schools with limited access to knowledgeable healthcare professionals to assist them (Leclair, 2018).

Sharing day-to-day mobilization strategies for families coping with physical mobility challenges is part of the nurse and allied health professional role (Best, Routhier, & Miller, 2015; Powell-Cope, Pippins, & Young, 2017; Thurman, Harrison,

Garcia, & Sage, 2019). Given the significance of these mobilization activities for the OI families in our study, nurses, physiotherapists, and occupational therapists should regularly revisit OI families' needs for transportation and mobilization assistance. Caregivers and children with OI should continue to share their mobilization strategies with the clinical team allowing clinicians to offer additional feedback on these strategies and compile best practices (Powell-Cope et al., 2017).

OI medical services should be more accessible with OI specialists being easier to locate. OI caregivers shared that their day-to-day caregiving experiences were improved once they had access to the specialized OI care available at the study site. However, a few caregivers also shared that the time taken to travel to appointments, to complete surgeries, to communicate between healthcare teams about their child's needs, and to fill out reimbursement forms were all too time-consuming and complicated. An interdisciplinary clinical team at the study site has worked to develop a passport and a transfer summary tool to help families of young adults with OI better coordinate the transition from paediatric to adult healthcare services, helping to support continuity of care (Jeong et al., 2019). Institutions and policymakers should work to ensure that all medical services are accessible, efficient, easy to navigate, with as little family separation as possible, and with continuity of care.

One way of making OI services more accessible is through digital health initiatives. Nurses and clinical researchers should ensure that they are sharing accessible online resources for families coping with rare conditions like OI. For families coping with rare and relatively unknown conditions, the Internet may be one of their only sources of information (DeHoff, Staten, Rodgers, & Denne, 2016; Knapp, Madden, Wang, Sloyer, & Shenkman, 2011). Clinicians caring for families of children with other rare conditions, such as cerebral palsy, have worked to develop portable and/or online

1 services to provide easier and reputable access to specialized supports for the
2 management of day-to-day care (Camden et al., 2019). Internet-based support tools may
3 be particularly useful for supporting the day-to-day care of children with unique needs
4 such as OI - particularly among families who do not live near specialist medical centres,
5 or who do not have acquaintances living with the condition (Castro et al., 2019). The
6 study site recently adopted the *Upopolis* platform, which is a secure social network
7 which helps children and families with medical support needs to connect with each
8 other and share resources ("Upopolis Launches at Shriners," 2017). The ongoing
9 development of such tools for OI families to learn about and share resources regarding
10 care needs may better support these families in the future, as well.

11 Considering the significance study participants placed on mobilization and
12 transportation in day-to-day care, clinical teams might also work to develop virtual tools
13 to share mobilization and transportation strategies for OI families online, so that OI
14 families have access to these clinical supports at home (Powell-Cope et al., 2017).
15 Clinicians have developed telehealth programs to help improve access to specialist
16 resources for families coping with other rare conditions (Camden et al., 2019). OI
17 clinical sites might also develop telehealth programs to provide day-to-day clinical
18 support for mobilization, fractures, pain management, and community health referrals,
19 so that families who live further from the study site have easier access to clinical
20 expertise. Such resources could further support caregivers in helping their children to
21 thrive with OI.

22 Policymakers should focus on building supports for trusted respite care and
23 other high-quality services. Policymakers should work to build more comprehensive
24 and qualified respite support services for caregivers of care recipients with complex
25 needs. A few caregivers in this study suggested that childcare services provided by

specialized treatment centres, such as special drop-off days or summer camps, would be appreciated. Another option might be to develop OI caregiving teaching materials to help caregivers teach their friends and extended family on how to care for their child day-to-day. A few of the caregivers mentioned that even when they had community supports like adapted buses and wheelchair ramps, those services often did not meet their unique caregiving needs, so they did not use them. Policymakers should work to ensure that such services are adapted and truly accessible to families with complex care needs, so that these families can learn to trust and rely on the services developed for them.

What does this paper contribute to the wider global clinical community?

- These results showcase what being a caregiver for a child with a rare and chronic physical condition - Osteogenesis Imperfecta - involves on a day-to-day basis.
- Caregivers of children with rare and chronic physical conditions like OI have unique needs, such as physical mobilization support needs, as well as needing better access to medical information, accessible schools, and appropriate care services.
- Nurses, clinicians, and policymakers who work with families coping with OI and similar conditions should develop interventions and policies that support the unique needs of these families; these interventions and policies should facilitate the long-term strategies that caregivers have already developed to support their day-to-day caregiving activities.

References

- Andrews, M. (2013). Measuring exertion during caregiving of children and young adults with cerebral palsy who require assistance for mobility and self-care. *Phys Occup Ther Pediatr*, 33(3), 300.
- Arabaci, L. B., Bozkurt, S., Vara, S., Ozen, S., Darcan, S., & Simsek, D. G. (2015). Difficulties experienced by caregivers of patients diagnosed with osteogenesis imperfecta (OI): Example of a hospital. *J Pak Med Assoc*, 65(7), 764-770.
- Bernehall, C. I., & Brodin, J. (2002). What families with children with brittle bones want to tell. *Child: Care, Health and Development*, 28(4), 309-315. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=emed8&NEW S=N&AN=34846908>
- <https://onlinelibrary.wiley.com/doi/abs/10.1046/j.1365-2214.2002.00282.x>
- Best, K., Routhier, F., & Miller, W. (2015). A description of manual wheelchair skills training: current practices in Canadian rehabilitation centers. *Disability and Rehabilitation: Assistive Technology*, 10(5), 393-400. doi:10.3109/17483107.2014.907367
- Bozkurt, S., Baysan Arabaci, L., Vara, S., Ozen, S., Goksen, D., & Darcan, S. (2014). The impact of psycho-educational training on the psychosocial adjustment of caregivers of osteogenesis imperfecta patients. *J Clin Res Pediatr Endocrinol*, 6(2), 84-92. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/24932601>
- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qual Res Psychol*, 3(2), 77-101. doi:doi: 10.1191/1478088706qp063oa
- Camden, C., Pratte, G., Fallon, F., Couture, M., Berbari, J., & Tousignant, M. (2019). Diversity of practices in telerehabilitation for children with disabilities and effective intervention characteristics: results from a systematic review. *Disability and rehabilitation*, 1-13. doi:10.1080/09638288.2019.1595750
- Castro, A. R., Chougui, K., Bilodeau, C., & Tsimicalis, A. (2019). Exploring the Views of Osteogenesis Imperfecta Caregivers on Internet-Based Technologies: Qualitative Descriptive Study. *Journal of Medical Internet Research*, 21(12), e15924.
- Chougui, K., Addab, S., Palomo, T., Morin, S. N., Veilleux, L. N., Bernstein, M., . . . Tsimicalis, A. (2020). Clinical manifestations of osteogenesis imperfecta in adulthood: An integrative review of quantitative studies and case reports. *American Journal of Medical Genetics Part A*.
- Collins, A., Hennessy-Anderson, N., Hosking, S., Hynson, J., Remedios, C., & Thomas, K. (2016). Lived experiences of parents caring for a child with a life-limiting condition in Australia: A qualitative study. *Palliat Med*, 30(10), 950-959.
- DeHoff, B. A., Staten, L. K., Rodgers, R. C., & Denne, S. C. (2016). The Role of Online Social Support in Supporting and Educating Parents of Young Children With Special Health Care Needs in the United States: A Scoping Review. *J Med Internet Res*, 18(12), e333. doi:doi: 10.2196/jmir.6722
- Dogba, M. J., Bedos, C., Durigova, M., Montpetit, K., Wong, T., Glorieux, F. H., & Rauch, F. (2013). The impact of severe osteogenesis imperfecta on the lives of young patients and their parents - a qualitative analysis. *BMC Pediatr*, 13(1), 153. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/24074180>
- Dogba, M. J., Rauch, F., Tre, G., Glorieux, F. H., & Bedos, C. (2014). Shaping and managing the course of a child's disease: parental experiences with osteogenesis

- imperfecta. *Disability & Health Journal*, 7(3), 343-349. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/24947576>
- Hill, C. L., Baird, W. O., & Walters, S. J. (2014). Quality of life in children and adolescents with Osteogenesis Imperfecta: a qualitative interview based study. *Health & Quality of Life Outcomes*, 12(1), 54. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/24742068>
- Jeong, S., Chougui, K., Mercier, C., Wong, T., Lafrance, M. E., Gagnon, V., . . . Tsimicalis, A. (2019). Development of the Good2Go MyHealth Passport for individuals with Osteogenesis Imperfecta: A knowledge-synthesis study. *Int J Orthop Trauma Nurs*, 33, 27-34. doi:doi: 10.1016/j.ijotn.2018.11.005
- Knapp, C., Madden, V., Wang, H., Sloyer, P., & Shenkman, E. (2011). Internet use and eHealth literacy of low-income parents whose children have special health care needs. *J Med Internet Res*, 13(3), e75. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/21960017>
- Leclair, A. (Producer). (2018, 2018/08/25). EXCLUSIVE: Saskatchewan boy creates splint kit for fellow patients at Shriners Hospital for Children. Retrieved from <https://globalnews.ca/news/4068922/exclusive-saskatchewan-boy-creates-splint-kit-for-fellow-patients-at-montreal-shiners-hospital/>
- Lincoln, Y. S., & Guba, E. G. (1985). Naturalistic inquiry. Newberry Park. In: CA: Sage.
- Luijckx, J., Putten, A. A. J., & Vlaskamp, C. (2017). Time use of parents raising children with severe or profound intellectual and multiple disabilities. *Child: Care Health Dev*, 43(4), 518-526. doi:10.1111/cch.12446
- Marini, J. C., Forlino, A., Bachinger, H. P., Bishop, N. J., Byers, P. H., Paepe, A., . . . Semler, O. (2017). Osteogenesis imperfecta. *Nat Rev Dis Primers*, 3, 17052. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/28820180>
- McCann, D., Bull, R., & Winzenberg, T. (2012). The daily patterns of time use for parents of children with complex needs: A systematic review. *J Child Health Care*, 16(1), 26-52.
- Pelentsov, L. J., Laws, T. A., & Esterman, A. J. (2015). The supportive care needs of parents caring for a child with a rare disease: A scoping review. *Disability & Health Journal*, 8(4), 475-491. doi:<https://doi.org/10.1016/j.dhjo.2015.03.009>
- Polit, D., & Beck, C. (2012). *Nursing research: Generating and assessing evidence for nursing practice* (9 ed.). New York: Wolter Kuwer Health - Lippincott Williams & Wilkins.
- Powell-Cope, G., Pippins, K., & Young, H. (2017). Teaching Family Caregivers to Assist Safely with Mobility. *AJN The American Journal of Nursing*, 117(12), 49-53. doi:10.1097/01.NAJ.0000527485.94115.7e
- Samia, L. W., O'Sullivan, A., Fallon, K. C., Aboueissa, A.-M., & Hepburn, K. W. (2018). Building on self-efficacy for experienced family caregivers: The Savvy Advanced Program. *The Gerontologist*. doi:10.1093/geront/gny016
- Sandelowski, M. (1995). Sample size in qualitative research. *Research in Nursing & Health*, 18(2), 179-183.
- Sandelowski, M. (2000). Whatever happened to qualitative description? *Res Nurs Health*, 23(4), 334-340. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/10940958>
- Santos, M. C. D., Pires, A. F., Soares, K., & Barros, L. (2017). Family experience with osteogenesis imperfecta type 1: the most distressing situations. *Disabil Rehabil*, 1-7. doi:<https://dx.doi.org/10.1080/09638288.2017.1334236>

- 1 Szczepaniak-Kubat, A., Kurnatowska, O., Jakubowska-Pietkiewicz, E., & Chlebna-
2 Sokol, D. (2012). Assessment of quality of life of parents of children with
3 osteogenesis. *Adv Clin Exp Med*, 21(1), 99-104. Retrieved from
4 [http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=emed14&NE](http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=emed14&NEWS=N&AN=364542406)
5 [WS=N&AN=364542406](http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=emed14&NEWS=N&AN=364542406)
- 6 Thurman, W., Harrison, T., Garcia, A., & Sage, W. (2019). The social construction of
7 disability and the capabilities approach: Implications for nursing. *Nursing*
8 *Forum*, 54(4), 642-649. doi:10.1111/nuf.12389
- 9 Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting
10 qualitative research (COREQ): a 32-item checklist for interviews and focus
11 groups. *Int J Qual Health Care*, 19(6), 349-357. Retrieved from
12 <https://www.ncbi.nlm.nih.gov/pubmed/17872937>
- 13 Trejo, P., & Rauch, F. (2016). Osteogenesis imperfecta in children and adolescents-new
14 developments in diagnosis and treatment. *Osteoporos Int*, 27(12), 3427-3437.
15 Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/27492436>
- 16 Upopolis Launches at Shriners. (2017). Retrieved from
17 [https://www.kidshealthlinks.org/blog/2017/12/20/eq6nm37otnyd742ymmyf35fv](https://www.kidshealthlinks.org/blog/2017/12/20/eq6nm37otnyd742ymmyf35fv0xpa51)
18 [0xpa51](https://www.kidshealthlinks.org/blog/2017/12/20/eq6nm37otnyd742ymmyf35fv0xpa51)
- 19 Vanz, A. P., Felix, T. M., da Rocha, N. S., & Schwartz, I. V. (2015). Quality of life in
20 caregivers of children and adolescents with Osteogenesis Imperfecta. *Health &*
21 *Quality of Life Outcomes*, 13, 41. doi:[http://dx.doi.org/10.1186/s12955-015-](http://dx.doi.org/10.1186/s12955-015-0226-4)
22 [0226-4](http://dx.doi.org/10.1186/s12955-015-0226-4)

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24

Tables

Table 1. Key interview questions and prompts related to day-to-day care.

<i>Main questions:</i>
1) Can you describe what a regular day looks like when caring for your child with OI?
2) And then can you describe a more challenging day recently?
<i>Prompts:</i>
What are some of the duties or responsibilities of caring for a child with OI?
How has your caring for your child changed over time?
What areas of your family's life do you feel are going well?
What are some areas that you would like to see improve?
What happens on a day with a bone break? What happens in the days that follow?
Do you feel you know about community resources for families of children with chronic conditions?
How has caregiving affected your lifestyle and schedule? Your job? Your relationships? Your personal care?
How do you feel about your social support?
Do you have access to extra help and/or care around the house?

1 Table 2. Demographic data.

<u>Demographic Trait</u>	<u>n (individuals, unless otherwise indicated)</u>
Individuals interviewed	18
Number of families represented	14
Language of interview (as caregiver preferred)	
English	12
French	6
Median caregiver age in years (range)	37.5 (24-57)
Caregiver's sex (/18 caregivers)	
Female	13
Male	5
Marital Status (/18 caregivers)	
Married or common-law	14
Single (never married)	2
Separated or divorced	2
Residential region (/14 families)	
Local (< 2 hours' drive from Montreal)	7
Within Quebec, not local	1
Other Canadian region	3
International	3
Highest level of education (/18 caregivers)	
Some post-secondary (university or college)	5
Received university or college degree/diploma	11
Postgraduate	2
Estimated family income (/14 families)	
Less than \$25 000	3
\$25 000 - \$50 000	1
\$50 000 - \$80 000	1
More than \$80 000	7
Do not know	1
Prefers not to answer	1
Relationship to child with OI (/18 caregivers)	
Mother figure (biological or guardian)	13
Father figure	5
Child's OI type (/14 families)	
I	3
III	4
IV	6
VI	1

Number of families with more than 1 child with OI in the household	3
Ages of children with OI (17 children with OI)	
Baby (0-12 months)	2
Toddler (13 months - 3 years old)	1
Pre-school (4-5 years old)	1
School-aged (6-12 years old)	11
Teenager (13-18 years old)	2
How would you describe the health of your child with OI right now? (/18 caregivers)	
Excellent	5
Very good	4
Good	8
Fair	1
Poor	0
Family history of OI (/14 families)	
Yes	3
No	11

Note: This table has been adapted for this report from the demographic data table portrayed in (Castro et al., 2019). The two tables are similar but not identical.