
BRIEF REPORT

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The Direct and Indirect Financial Costs Sustained by Parents of Children with Osteogenesis Imperfecta: A Brief Report

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ABSTRACT

Osteogenesis Imperfecta (OI), also known as brittle bone disease, is a rare genetic condition characterized by increased bone fragility. The financial costs of caring for a child with a rare condition can be a significant source of stress for families. As such, the financial costs of care are a concern to the clinicians who care for these families. However, the financial impact of caring for a child with OI is under-reported. A secondary analysis was conducted on data collected from a qualitative study in nursing. The aim was to offer preliminary insights into the financial impacts of caring for a child living with OI. Participants reported many direct out-of-pocket costs, which presented with reimbursement barriers. Other indirect costs resulted from missed work, career changes, or workforce departures. Policy and clinical implications include optimizing accessibility of supportive childcare policies, simplifying healthcare reimbursements, and regularly assessing families' knowledge of available financial supports.



KEYWORDS

Cost of Illness, Nursing Research, Caregivers, Rare Diseases, Parenting

1 | INTRODUCTION

Osteogenesis Imperfecta (OI) is a rare genetic condition that primarily results in increased bone fragility. (1) The financial costs of caring for a child with a rare condition can be an important source of stress for families. These stressors are also of concern to the clinicians responsible for providing these families with holistic care. (2) Yet, the costs incurred from caring for a child living with OI remain understudied. (3) This report seeks to offer preliminary insights into the financial impacts of caring for a child living with OI.

2 | METHODS

Following ethical approval (A05-B39-17B, McGill University Faculty of Medicine), a secondary analysis of qualitative data was conducted. The financial dataset was derived from a primary nursing science study conducted with OI caregivers at a paediatric hospital in Montreal, Canada. (4) Purposive sampling was used to recruit caregivers when they arrived at the study site for a scheduled OI appointment. Participants provided informed, written consent and were interviewed. Demographic data were analyzed descriptively, and audio-recorded interviews were transcribed and thematically analyzed according to direct and indirect financial costs. (5, 6)

3 | RESULTS

Eighteen parents of children with OI from 14 families participated. Demographic data are listed in **Table 1**. Families shared the direct and indirect costs of caring for children with OI. The direct costs were financial expenditures incurred by families. (6) Indirect financial costs included productivity losses due to time spent trying to access reimbursements; as well as income losses resulting from the career consequences of caregiving. **Table 2** displays quotes exemplifying each sub-theme.

3.1 | Out-of-Pocket Costs

Varying direct costs were incurred from caring for a child with OI. One family estimated their yearly OI expenses, including travel and other items, averaged \$5,000; whereas another family's estimate was lower, amounting to \$1,000-\$1,500 per year. Four families were saving to buy equipment to optimize their child's mobility, including the use of a lighter, more travel-friendly wheelchair, and the installation of home stair-lifts. Building a wheelchair ramp and installing handrails cost one family nearly \$500. A few families expressed concerns regarding dental costs resulting from the negative effects of OI on jawbone and teeth development (1). One mother estimated her daughter's various orthodontic treatments, such as braces due to her many missing adult teeth, will ultimately cost her between \$15,000 to \$20,000, because their provincial healthcare plan does not cover orthodontic treatments.

3.2 | Navigating Administrative Processes for Reimbursements

Parents explained that healthcare systems are complicated, and OI families have to navigate the healthcare system, and request more reimbursements, compared to families without rare orthopedic conditions. Some out-of-pocket expenses were reimbursable from the families' health insurance plans, but reimbursement processes were time-consuming. One out-of-province parent estimated that for every hospital appointment, nearly \$1,000 in travel-related out-of-pocket expenses were incurred upfront, which could only be reimbursed after submitting a burdensome application. Only by chance did another family learn of their eligibility for medical tax deductions. For smaller expenses, some families opted to absorb their out-of-pocket expenses, rather than submitting their claims.

Demographic Trait	Number (n)
Individuals interviewed	18
Number of families represented	14
Median caregiver age in years (range)	37.5 (24-57)
Parental status (/18 caregivers)	
Mother figure	13
Father figure	5
Marital status (/18 caregivers)	
Married or common-law	14
Single (never married)	2
Separated or divorced	2
Residential region (/14 families)	
Quebec	8
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 CAD	3
\$25,000 - \$50,000 CAD	1
\$50,000 - \$80,000 CAD	1
More than \$80,000 CAD	7
Do not know	1
Prefers not to answer	1
Ages of children with OI ¹ (n=17)	
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

TABLE 1 Demographic Data¹OI = Osteogenesis Imperfecta

Sub-Theme	Participant	Exemplar Quote
Out-of-Pocket Costs	Family 9, Mother figure to a pre-school child with Type III OI, Lives in Quebec	"I can tell you for a [second] wheelchair, the wheelchair we're looking at for [child with OI] right now costs \$4,000. And we think that's a good fit. I mean, that brand of wheelchair is a good fit for him based on what we found out about other kids with the same type and his size and his therapy and all of that. But it's \$4,000 . . . [which provincial insurance will not cover] unless you qualify for the exception."
Navigating Administrative Processes for Reimbursements	Family 8, Mother figure to a school-aged child with Type VI OI, Lives in Canada outside of Quebec	"So, let's say they call, the doctor wants to see [child with OI]. So, the government will pay for our [airplane] tickets to [the nearest city with specialized pediatric services], because they're about \$2,000 each, so the government pays for that. But our hotel and our meals, we need to pay for out-of-pocket, and then submit our receipts, and get reimbursed. That doesn't always happen in a timely fashion. So, we are probably waiting on about \$8,000 of reimbursements, probably going back to 2014."
Income Losses due to Career Effects of OI Caregiving	Family 13, Mother figure to a school-aged child with Type III OI, Lives in Canada outside of Quebec	"He [child with OI] was looking at rodding surgeries, and he was looking at . . . we were coming out to Montreal every seven weeks, and usually for a week at a time. There's no – my job would not give me the time."

TABLE 2 Participant Quotes Exemplifying the Sub-Themes for Direct and Indirect Financial Costs to Families of Children Living with OI¹

¹OI = Osteogenesis Imperfecta

3.3 | Income Losses due to Career Effects of Caregiving

Parents relayed how caring for a child with OI could lead to missed paid work time and complications with work schedules. For one family, if their child sustained a fracture, the child needed to be flown to a hospital situated several hours away. Such events could result in several days or even weeks of missed work for the accompanying parent, depending on the number of treatments and surgeries needed. Another father used up his vacation time to attend his child's healthcare appointments. Some of the career challenges experienced by parents

were mitigated by supportive employers and flexible work opportunities. Other parents changed their career plans, opting for greater work flexibility or leaving the paid workforce altogether. Six of the seven parents who described altering their career plans due to their children's OI diagnoses were mother figures. These mothers either: (1) chose less desirable but more flexible job positions (n=2); (2) chose to become part-time or casual employees, rather than maintain their full-time statuses (n=1); or (3) left the paid workforce all together (n=3).

4 | DISCUSSION

This report offers preliminary insights into the financial impacts of caring for a child with OI. Participants in our study incurred varying direct out-of-pocket expenses. These costs are similar to those incurred by families caring for children with other complex care needs, particularly out-of-pocket costs relating to travel and equipment expenses. (7) Accessing reimbursements was also a source of financial difficulty for families. Andersen et al. (8) report that families of children with disabilities have a high likelihood of not being reimbursed for expenses; our findings echo this conclusion.

Families experienced income losses due to career re-orientation and/or leaving the paid workforce. The unpredictability of fractures and accompanying surgeries often led to hospitalisations of variable length and occurrence, affecting parents' abilities to remain employed. These results are consistent with pediatric oncology research performed by Warner et al. (9) which reports that families' incomes suffer the most when their children's care needs are less predictable. Additionally, mothers of children with OI were primarily impacted by reduced employment. This gendered phenomenon is common in the management of childhood chronic illnesses, where the care work is disproportionately borne by mothers. (2, 7)

Policymakers should investigate ways to mitigate the gendered financial effects of caring for a child with a rare condition, potentially by establishing more accessible childcare services for children with disabilities, encouraging telework options, and offering paid parental caregiver stipends. Healthcare institutions should ensure that medical teams avoid changing surgery/treatment schedules as much as possible. Institutions should work to support simplified reimbursement strategies. Finally, nurses and allied health clinicians should regularly assess OI families' knowledge of and need for available financial support.

4.1 | Limitations

The sample had limited representation from young caregivers, single-income households, and low-income families. The scope of inquiry related to costs and financial impacts was limited in the original study.

5 | CONCLUSIONS

The financial burden associated with caring for children with rare diseases is a critical, but often overlooked, component of the patient and family healthcare experience. (2, 8) This study offers important insights for clinicians, institutions, and policymakers to better understand and mitigate the family-incurred financial costs resulting from the care needs of children living with the rare condition of OI.

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