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Nursing:
The Heart of Healthcare

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Cover art by: Claire Chabot, 1st Place Winner of 2021 MJM Cover Competition

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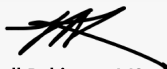
Letter from the Editors: The Changing Face of Healthcare and Scholarship

The McGill Journal of Medicine (MJM) was founded in the 1940s and then relaunched numerous times afterwards with the intent of providing medical and graduate students with opportunities to publish their first articles, thus offering a stepping stone into the broader world of academic scholarship.

While the MJM's dedication to publishing student work has unwavered throughout the years, much has changed since the inception of the MJM. The reality of healthcare in the 21st century has shifted dramatically towards inter-professional care and interdisciplinary research efforts. In fact, the McGill Faculty of Medicine recently underwent a renaissance in 2020 and changed their name to the McGill Faculty of Medicine and Health Sciences to highlight the increasing importance of collaborative care across different healthcare professions to patient care and research. In this new digital era with an explosion of new research articles being published each year and instantaneous access to research from around the world, emphasis on research and scholarship needs to become more prevalent at the undergraduate level not only in medicine, but also all the other schools and departments in the Faculty. Efforts such as those by Dr. Argerie Tsimicalis to encourage scholarship and critical research appraisal in undergraduate healthcare programs that are traditionally more "clinically oriented" are essential to producing a new generation of healthcare professionals equipped to take on the deluge of research in the digital era. These skills are crucial to filter high quality, relevant research and translate these findings into new practises and policies to enhance treatments and further existing research in all domains of patient care.

In light of this evolving face of healthcare, it became increasingly apparent to us on the Editorial Board of the MJM that our scope had to be broadened beyond the medical and graduate student population that we traditionally serve. As our first foray into inter-professional special issues, it is our hope that *Nursing: The Heart of Healthcare* will encourage research in our allied health partners and inspire further cross disciplinary collaborations. While we may come from different training backgrounds, we remain united in our goal of providing the best care possible for our patients.

Sincerely,



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Foreword: MJM's Inaugural Nursing Issue

Charged with the reality that most undergraduate nursing students are interested in building their “clinical skills” rather than learning about research, I am tasked with persuading these students to embrace research into their practices. I need to solicit their buy-in, engage them in the process of learning a new language, and hopefully instill a life-long commitment to use research findings, despite all the barriers, as one of their sources of knowledge guiding their practices. The reality of COVID-19 showcased the need for more research-savvy nurses who can critically appraise research and translate these findings into practice. However, the bulk of our nursing profession is prepared at the baccalaureate level in Canada and worldwide. Many of the skill sets move beyond foundational knowledge and are only learnt at the graduate level, reinforcing the need to strengthen the pipeline of recruiting and training nurses at the doctoral and postdoctoral level. Similar to other disciplines, this process needs to begin at the baccalaureate level by exposing students to a range of research opportunities, offering research practicums, and immersing them in research communities. If not, I fear that I and our nursing profession may contribute to unsafe care, poor patient outcomes, harmful policies and standards, and a dismal transformation of our complex health care systems. And yet, there remains a strong untapped potential in the training of undergraduate nursing students. Having trained and published a handful of undergraduate nursing students, there are ways to mobilize and circumvent existing barriers. The process may begin with introducing novel pedagogical approaches to teaching research in nursing education, creating new student partnerships, and recognizing the exceptional contributions of nursing across educational preparations.

Novel ways are needed to teach undergraduate nursing students about research. The guiding principle is that the more interactive and experiential the learning strategies used, the more likely students will be motivated to learn about research. (1) While these principles may be easier to apply one-on-one, varying challenges with few notable successes published, preclude *infecting* students with the love of research and *inoculating* them against an aversion to research. (2, 3) Spires et al. (1) showcased the use of games, cross word puzzles, and analysis of common multimedia data may be some strategies used to teach research in large classroom settings. Personally, my efforts to *infect* undergraduate students led to the creation of a new group assignment by partnering with (The McGill Journal of Medicine) (MJM), a student-led, open access, journal, to operationalize the research domain standard of the Canadian Association of Schools of Nursing (CASN), which entails to “foster the development of critical thinking and research abilities to use evidence to inform nursing practice”. (4)

The onset of COVID-19 sparked the need to convert a historically didactic, 3-credit, research course to be delivered remotely, asynchronously and experientially to 164 undergraduate nursing students. As explained in my blog (5), efforts to deliver this course “remotely” and “asynchronously” warranted the inclusion of an online group assignment. Inspired by the journal “Evidence-Based Nursing” and with the full support of the McGill Journal of Medicine, a four-part assignment was collaboratively created with input from faculty and former nursing student, Ms. Kayla Douglas. The assignment was called “Influencing Nursing: Collaborative Creation of an Expert Commentary, Infographic and Tweet”, and available on the MJM website for others interested to use in their teaching. The assignment sought to

train undergraduate nursing students how to critically review a research article by drafting a succinct expert commentary derived from research conducted by their nursing professors. A subsequent part of the assignment aimed at optimizing students capacity for translating research into practice. Students learned to convey key messages of their commentary into a succinct, understandable and creative infographic that would target audiences with varying health literary skills. Subsequently, students reflected on the use of social media for disseminating evidence by creating a Tweet as part of their commentary. Together, these strategies were designed to help students move beyond passive ways of disseminating knowledge and to think about how to incorporate innovative means to craft the message, package the information, and share the research findings. The final part of the assignment arose half-way during course delivery when MJM created a Special Nursing Issue and welcomed the students' assignments as part of their call for papers. The special issue was part of our collective efforts to celebrate the 2020 Year of the Nurse and Midwife showcasing how students may influence nursing at the baccalaureate level. Students were offered a 2 per cent bonus mark if they submitted their final assignment to MJM; prompting them to learn about the peer review process and the International Committee of Medical Journal Editors authorship guidelines.

The pathway towards advancing nursing scholarship is not without its pitfalls. Students were expected to achieve the highest level of learning by creating and innovating. I hope a few skills were learnt, better positioning them for the transformation of our complex health care systems. In this inaugural nursing issue, myself and the MJM editorial team wish to showcase the contributions nursing offers from a baccalaureate student to a doctoral-prepared nursing scientist.

Argerie Tsimicalis, RN PhD

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Acute Cardiovascular Effects of Vaping Compared to Cigarette Smoking in Young Adults

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ABSTRACT

Background: First introduced as the safer alternative to smoking, vaping has become a popular activity among young adults. However, little is known about its potential health effects. This pilot project examined the acute cardiovascular effects of nicotine vapes/e cigarettes (EC) compared to tobacco cigarettes (TC) in young adults to determine if vaping is more detrimental to cardiovascular health than traditional cigarettes.

Methods: 16 healthy participants (7 M, 9 F; 20.2 ± 1.9 years) were recruited to participate in the study. Anthropometric measures were determined upon entry into the study. In addition, circulatory measures (heart rate [HR], blood pressure [BP] and heart rate variability [HRV]) were measured prior to and 10-min following vaping or cigarette smoking and in response to an orthostatic challenge.

Results: Resting circulatory and HRV measures were not different between chronic EC-users and TC-smokers. Both vaping and cigarette smoking-induced a significant increase in cardiovascular measures (HR and BP) but not HRV measures. Both groups responded similarly to the orthostatic challenge prior to and following vaping/smoking.

Conclusion: These results indicate that, from a cardiovascular perspective, vaping induces similar acute effects as cigarette smoking and that young adults should be counselled about these adverse effects accordingly.

KEYWORDS

Cigarettes, Vapes, Nicotine, Cardiovascular system, Adolescents

1 | INTRODUCTION

A substantial amount of literature indicates that tobacco cigarette (TC) smoking is related to a plethora of cardiovascular diseases (1); causing approximately 30% of car-

diovascular disease related deaths. (2) Smoking TC has been noted to increase an individual's chances of developing atherosclerotic diseases such as angina, acute coronary syndrome, stroke and sudden death. (3) Tobacco cigarettes contain over 7000 chemical compo-

nents, including nicotine (with concentrations of 1.99 ± 0.20 mg/cigarette), tar, carbon monoxide, acrolein and pro-oxidants. (4) Nicotine has been shown to be a predominant factor of accelerated atherogenesis and cardiovascular disease. (5)

Electronic cigarettes (e-cigarettes, EC) were introduced in 2006 as a more health-conscious alternative to cigarette smoking. (6) EC are electronic heating devices which create an aerosolized mixture of liquid containing stimulants (e.g., nicotine, marijuana), flavoring and solvents for heating that can be inhaled. (7) Nicotine concentrations in EC can range from 1.6 - 19 mg per cartridge. (8) The adverse effects of chronic EC use include bronchitis, emphysema, respiratory tract irritation and cardiovascular disease. (2)

It has been well established that the acute effects of smoking TC can change heart rate variability (HRV) parameters, leading to an increase in the low-frequency to high frequency (LF/HF) ratio. (9) This increase is attributable primarily to the effects of nicotine. (10) Individuals who smoke cigarettes regularly often experience a reduction in HRV, indicating an increased sympathetic nervous system (SNS) activity, increased heart rate (HR) and blood pressure (BP). (11) Similarly, vaping EC that contain nicotine has also been found to induce an increase in SNS activity. (10, 12)

While vaping is gaining popularity (4), little research has been done to determine its possible cardiovascular side effects (2) and minimal research has compared the effects of smoking tobacco cigarettes (TC) with vaping EC on HRV. (10) HRV is a reliable, non-invasive tool for determining autonomic nervous system control of the heart. In a study of 100 smokers (42 TC smokers and 58 chronic EC users), Arastoo et al (10) found that baseline cardiovascular and HRV measures were similar between TC-smokers and EC-users and that HR, systolic blood pressure [SBP], diastolic blood pressure [DBP], and mean arterial pressure [MAP] significantly increased following acute exposure to TC and EC. However, HRV measures were not altered. Interestingly, the increase in blood pressure [BP] was significantly greater in the TC smokers, which the authors believe may have been due to the greater number of chemicals contained in TC. So far,

other studies that have compared the acute autonomic effects of smoking TC with EC-use have examined HR, SBP and DBP as surrogate measures. (12) HR has been consistently shown to increase, whereas contradictory results have been reported for SBP and/or DBP. Similar to the work of Arastoo et al (10) a few studies have demonstrated that the cardiovascular autonomic effects of smoking TC are greater than vaping EC.

Moreover, no studies have examined the dynamic influence of a sympathetic maneuver (such as an orthostatic challenge) on HRV in TC-smokers and EC-users. One study with TC smokers examined the dynamic influence of parasympathetic (PNS) (controlled breathing) and sympathetic (SNS) maneuvers (hand-grip exercise) on HRV (13). Barutcu et al (13) found that parasympathetic (vagal) modulation was blunted in smokers during a controlled breathing exercise.

Therefore, the purpose of this study is to examine the acute (short-term) cardiovascular effects of cigarette smoking (TC) and e-cigarettes (EC). This research is important as EC are becoming increasingly popular among cigarette smokers and those who have no previous history of smoking, yet, there are gaps in the literature examining the acute health consequences of regular EC use. (2) This project examined the acute cardiovascular effects of smoking TC in young adults in comparison to those who vape EC as well as their response to a postural challenge. Based upon our review of the literature, we hypothesized that there would be no difference in resting circulatory and HRV parameters between TC smokers and EC-users that heart rate and blood pressure would significantly increase following cigarette-smoking and vaping without measurable changes in HRV and that both groups would respond similarly to an orthostatic challenge. Since it was reported that that the perturbation in circulatory measures tends to be greater for individuals who use TC vs EC (10), we predicted that individuals in the TC group would have a higher HR, blood pressure, and increased sympathetic tone compared to individuals who vape EC.

2 | METHODS

This study used a pre-test, post-test cross-sectional design to assess the cardiovascular effects of smoking versus vaping. A flow chart representing the experimental design and outcome is depicted in **Figure 1**. The study was conducted between the months of January and March of 2020 and was approved by Trent University's Research Ethics Board (File 26045) in accordance with the Declaration of Helsinki.

2.1 | Participants

Participants were recruited through announcements made in class and posters placed at a post-secondary institution in Ontario, Canada. Interested participants contacted the research team. Exclusion criterion included the presence of known disease (e.g., cardiorespiratory, neurological, gastrointestinal, metabolic and psychiatric). Nineteen participants who either smoked traditional TC or used nicotine EC volunteered to participate in the study. Three participants were excluded from the study (one participant had a medical condition and two participants could not return for testing due to the COVID-19 pandemic lockdown). Participants ranged between the ages of 18 through 25 years and were assigned to their respective groups (TC vs EC) based upon their smoking history (a minimum of a 1-year smoking or vaping requirement for participation). The goal was to have exclusive smokers/vapers in the study, however, 4 smokers and 3 vapers disclosed that they occasionally vaped/smoked, respectively.

2.2 | Procedures

Participants visited the laboratory on two occasions. On the first visit, the experimental procedure was explained and written, informed consent was obtained. The participants then completed a questionnaire that was used to gather demographics and smoking/vaping history.

Anthropometric measures were taken including body mass, height, and skinfold measures. Body mass (kilogram [kg]) and height (metres [m]) were obtained us-

ing a Health-O-Meter scale (Health-O-Meter Corporation, Bedford Heights, Ohio). Body mass index (BMI) was determined by dividing the body mass (kg) by the participant's height (m^2). Body density was determined through skinfold measures, taken using a Harpenden skinfold caliper (FitSystems, Inc., Calgary, AB), from seven sites of the body (abdomen, biceps, thigh, iliac crest, midaxillary, chest, and subscapula) according to the Jackson-Pollock formulas for men and women. (14) Percent body fat was calculated from body density using the Siri body density conversion formula.

One week later, on the same day and time of the week, participants returned to the laboratory to have cardiovascular measures recorded. HR, SBP and DBP were determined in the supine and standing positions (prior to and following smoking/vaping) using an automated, non-invasive BP monitor (BpTRU) (Model BPM-300, VSM Medtech Ltd., Coquitlam, BC). Mean arterial pressure (MAP) and rate pressure product (RPP) were also determined. MAP, an indicator of the average blood pressure during one cardiac cycle was calculated from: $MAP = 1/3 (SBP-DBP) + DBP$. Rate pressure product (RPP), a measure of the workload of the heart, was determined from the product of heart rate and systolic blood pressure. Beat-by-beat R-R intervals were recorded continuously using a Polar Sport HR chest strap which transmitted the data to a wristwatch (Polar Vantage V2, Polar Sport, Montreal, QC).

For the supine condition, participants rested comfortably, in the supine position, on an examination table located in a quiet, light attenuated room. Participants refrained from speaking and moving for 10 min to allow for the recording of the cardiovascular measures. Cardiovascular measures were then recorded with the participant standing in an upright position for 10 minutes. Participants remained in the freestanding position without support for an adaptation period of 3 minutes followed by 10 minutes (or 512 heart cycles) of beat-by-beat data recording. Participants were reminded to relax, remain as still as possible, avoid talking and asked to refrain from leaning backwards on the examination table that was located behind them. Once the initial measures were obtained in the supine and standing po-

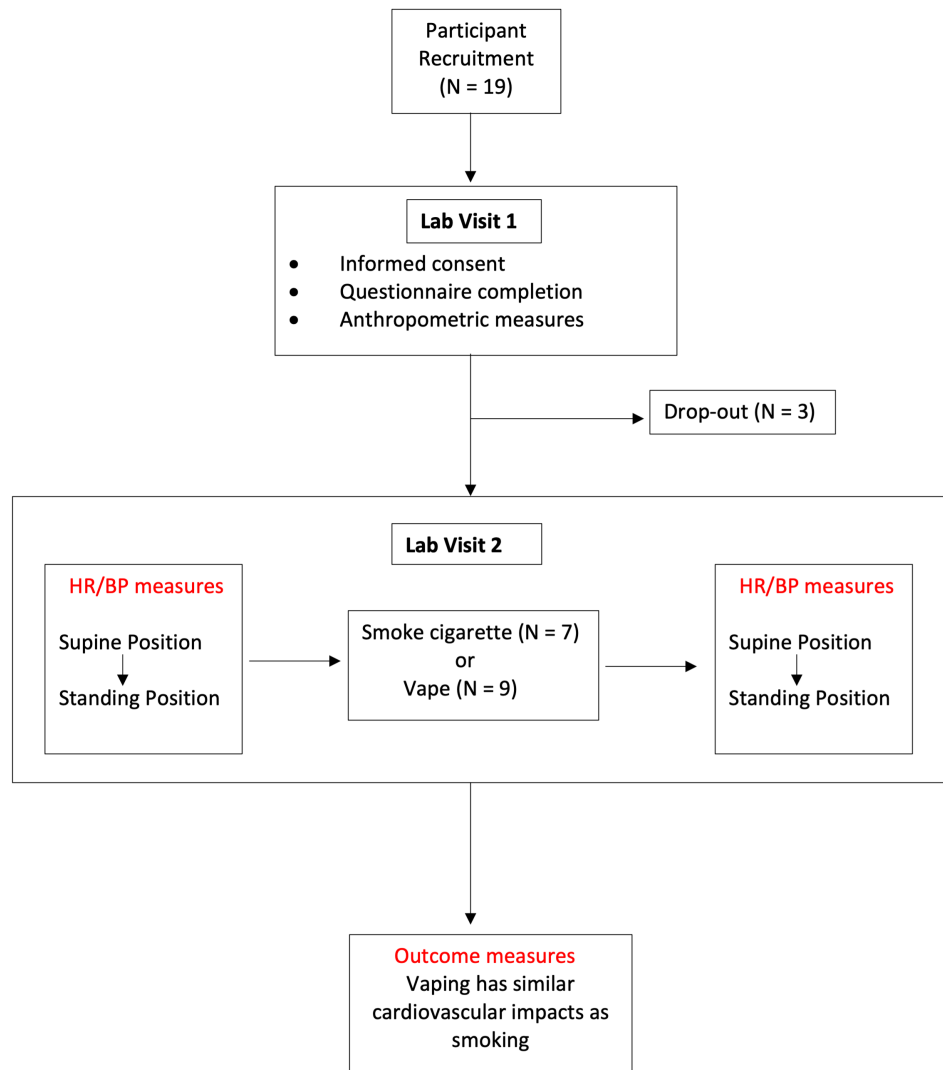


FIGURE 1 Study experimental design.

sitions, the participants were escorted to a designated smoking/vaping area. Participants were given a rest period of at least 3 minutes after reaching the smoking/vaping area so that their heart rate could return to baseline before smoking or vaping. The participants then either vaped e-cigarettes with 5.0% nicotine or smoked their normal cigarettes in the seated position for 2 and 5 minutes, respectively. Following this, participants returned to the laboratory to have their cardiovascular measures recorded again in the supine and standing positions. Polar Sport HR monitor recorded the R-R intervals. We determined HRV using Kubios (Kubios Oy, Kuopio, Finland).

2.3 | Statistical Analysis

Data were statistically analyzed using the computer program IBM SPSS Statistics for Macintosh, Version 27.0 (IBM Corp, Armonk, NY). Data are presented as means (\pm SD). Descriptive measures were assessed using an independent t-test or Chi-square test, whereas the hemodynamic and HRV variables of the TC smokers were compared to that of the EC users using a repeated measures ANOVA (within-group measure = time; between-group measure = TC vs EC). A p-value of 0.05 or less was considered significant.

3 | RESULTS

3.1 | Baseline characteristics of participants were similar

No significant differences were observed in baseline characteristics of the seven chronic TC smokers and the nine chronic EC-users, with the exception of years smoking or vaping (**Table 1**). Those individuals who smoked TC did so for a significantly ($p = 0.017$) longer period of time (4.7 years) compared to individuals who vaped EC (1.7 years). More than half (56%) of the participants were women, and they had a significantly ($p = 0.041$) higher BMI, compared to men, with an average BMI of 28.7 (9.3) versus 21.1 (2.33) kg/m². This pattern was more notable in the vaping (EC) group, albeit non-significant ($p = 0.066$), whereby BMI was 32.6 (10.5) for women compared to 20.9 (2.99) for men. In the group of TC smokers, BMI was 23.9 (5.4) for women versus 21.34 (1.7) for men.

3.2 | Cardiovascular responses were similar between cigarette smokers and vapers

The cardiovascular measures are presented in **Table 2**. There were no significant between-group (TC vs EC)

differences in the resting cardiovascular measures, response to smoking/vaping or to the orthostatic challenge.

Repeated measures ANOVA indicated that HR ($F(1,14) = 12.740$, $p = 0.003$), SBP ($F(1,14) = 16.980$, $p < 0.001$), DBP ($F(1,14) = 23.502$, $p < 0.001$), MAP ($F(1,14) = 29.797$, $p < 0.001$) and RPP ($F(1,14) = 30.028$, $p < 0.001$) significantly increased in response to smoking/vaping. Compared to the resting condition, HR ($F(1,14) = 382.164$, $p = < 0.001$), SBP ($F(1,14) = 11.070$, $p = < 0.001$), (DBP $F(1,14) = 55.746$, $p = < 0.001$), MAP ($F(1,14) = 55.304$, $p = < 0.001$) and RPP ($F(1,14) = 133.320$, $p = < 0.001$) were significantly increased in response to standing prior to smoking/vaping. Likewise, HR ($F(1,14) = 111.164$, $p = < 0.001$), SBP ($F(1,14) = 25.748$, $p = < 0.001$), DBP ($F(1,14) = 136.310$, $p = < 0.001$), MAP ($F(1,14) = 115.010$, $p = < 0.001$) and RPP ($F(1,14) = 161.092$, $p = < 0.001$) significantly increased in response to standing after smoking/vaping.

3.3 | HRV measures were significantly influenced by postural change

The HRV measures are presented in **Table 3**. No between-group (TC vs EC) differences were observed for the LF power, HF power, total power and the SNS in-

Variable	TC (n = 7)	EC (n = 9)	p value	ALL (n = 16)
Age (yrs)	21.1 (2.12)	19.4 (1.4)	0.075	20.2 (1.9)
Male sex (%)	3 (43)	4 (44)	0.949	7 (44)
Height (m)	1.7 (1.3)	1.8 (1.4)	0.528	1.7 (1.4)
Weight (kg)	66.5 (11.2)	82.3 (22.0)	0.087	75.4 (19.4)
BMI (kg/m ²)	22.8 (4.2)	27.4 (9.8)	0.235	25.4 (8.0)
Percent fat (%)	15.6 (11.8)	20.5 (10.8)	0.395	18.4 (11.2)
HR (bpm)	79.6 (9.8)	72.9 (13.5)	0.288	75.9 (12.1)
Systolic BP (mmHg)	108.8 (8.4)	109.7 (5.7)	0.794	109.3 (6.8)
Diastolic BP (mmHg)	72.2 (6.0)	71.9 (6.0)	0.910	72.0 (5.8)
Smoking/vaping (yrs)	4.7 (2.8)	1.7 (1.7)	0.017*	3.00 (2.66)
Smokes/vapes (/day)	3.7 (1.7)	4.0 (1.5)	0.727	3.88 (1.54)

Legend: Values are means \pm standard deviation (SD). BMI = body mass index, BP = blood pressure, HR = heart rate. * = $p < 0.05$

TABLE 1 Descriptive characteristics of participants at entry into the study.

Variable	Condition	Before smoking/vaping			After smoking/vaping		
		TC (n = 7)	EC (n = 9)	All (n = 16)	TC (n = 7)	EC (n = 9)	All (n = 16)
HR (bpm)	Rest	79.6 (9.8)	72.9 (13.5)	75.9 (12.1)	87.0 (13.1)	76.1 (14.6)	80.8 (14.6) **
	Stand	97.3 (14.8)	94.1 (13.8)	95.5 (13.8)*	101.4 (13.4)	94.8 (12.8)	97.7 (13.1)***
SBP (mmHg)	Rest	108.8 (8.4)	109.7 (5.7)	109.3 (6.8)	117.9 (8.0)	118.2 (8.6)	118.1 (8.1) **
	Stand	111.3 (7.8)	112.0 (5.7)	111.7 (6.5)*	114.4 (6.19)	119.1 (7.5)	117.0 (7.1)***
DBP (mmHg)	Rest	72.2 (6.0)	71.9 (6.0)	72.0 (5.8)	78.1 (6.9)	76.2 (7.4)	77.0 (7.0) **
	Stand	81.3 (4.3)	79.4 (6.5)	80.3 (5.6)*	83.5 (5.8)	84.5 (6.0)	84.0 (5.7)***
MAP (mmHg)	Rest	84.4 (6.0)	84.5 (5.5)	84.5 (5.6)	91.3 (6.0)	90.2 (7.1)	90.7 (6.5) **
	Stand	91.3 (5.1)	90.3 (5.9)	90.7 (5.4)*	93.8 (5.0)	96.0 (5.6)	95.0 (5.3)***
RPP (x 10 ³) (bpm.mmHg ⁻¹)	Rest	8.6 (1.2)	8.0 (1.7)	8.3 (1.5)	10.2 (1.5)	9.0 (2.0)	9.5 (1.8) **
	Stand	10.8 (1.6)	10.6 (1.8)	10.7 (1.7)*	11.6 (1.6)	11.3 (1.8)	11.4 (1.7)***

Legend: Values are means \pm (SD). Abbreviations are as follows: DBP = diastolic blood pressure; EC = electronic cigarette user; HR = Heart rate; SBP = systolic blood pressure; MAP = mean arterial blood pressure; RPP = rate pressure product; TC = tobacco cigarette smoker. * = $p < 0.05$ ("all" stand condition vs "all" rest condition - before smoking/vaping); ** $p < 0.05$ = ("all" rest condition after smoking/vaping vs "all" rest condition before smoking/vaping); *** $p < 0.05$ ("all" stand condition after smoking/vaping vs "all" rest condition before smoking/vaping).

TABLE 2 Circulatory responses of the participants.

indicator (LF/HF) in the resting condition and in response to smoking/vaping. There was a significant between-group effect for the PNS indicator (HF/total power) ($F(1,14) = 7.735$, $p < 0.015$) with the orthostatic challenge. Standing upright had a stronger PNS effect in the vaping group (both prior to and following vaping).

Repeated measures ANOVA indicated a significant main effect of standing on several autonomic indices. Compared to resting, both the HF ($F(1,14) = 5.936$, $p = < 0.029$) and PNS (HF/total) ($F(1,14) = 25.921$, $p = < 0.001$) indicator were significantly reduced with standing prior to smoking/vaping. Similarly, the HF ($F(1,14) = 8.047$, $p = < 0.013$) and PNS (HF/total) ($F(1,14) = 35.307$, $p = < 0.001$) indicator were significantly reduced with standing after smoking/vaping. In contrast, the SNS indicator was significantly increased in response to standing both before ($F(1,14) = 43.1$, $p < 0.001$) and after ($F(1,14) = 33.622$, $p < 0.001$) smoking/vaping.

4 | DISCUSSION

The major findings of this study are that resting hemodynamics and HRV measures are similar between chronic

TC smokers and EC vapers; HR and BP significantly increase in response to smoking and vaping, but HRV measures remain the same; and that the autonomic response to orthostatic challenge is not altered by smoking or vaping. These results support our hypotheses. However, our prediction that the circulatory responses of the TC-smokers would be greater was not observed.

To better comprehend the acute cardiovascular effects of smoking, HRV is often assessed. (11) HRV can be measured in both the time- and frequency-domain. Time domain analysis quantifies the R-R interval between two neighboring heartbeats over a set period of time. Frequency domain analysis is commonly used for analysis of brief recordings of heart rate (HR). (15) In a frequency-domain analysis, the variability in HR is put through a mathematical model (Fast-Fourier transformation) to provide indicators of autonomic function. High frequency (HF) power values are associated with parasympathetic activity, and low frequency power (LF) is correlated to both sympathetic and parasympathetic nervous system activity. (11) Parasympathetic nervous system (PNS) modulation can be inferred from the parasympathetic indicator (a ratio of the high frequency to total power), whereas sympathetic (SNS) modulation

Variable	Condition	Before smoking/vaping			After smoking/vaping		
		TC (n = 7)	EC (n = 9)	All (n = 16)	TC (n = 7)	EC (n = 9)	All (n = 16)
LF (ms ² /Hz)	Rest	824 (627)	2825 (5856)	1950 (4416)	403 (194)	2663 (5107)	1674 (3907)
	Stand	1466 (1681)	2740 (3364)	2182 (2755)	1658 (2388)	1979 (3348)	2182 (2755)
HF (ms ² /Hz)	Rest	617 (620)	1532 (1730)	1132 (1404)	394 (450)	2603 (4814)	1637 (3704)
	Stand	164 (154)	690 (1343)	460 (1022)*	162 (153)	496 (953)	350 (723)***
Total Power (ms ² /Hz)	Rest	2654 (1325)	5067 (7002)	4012 (5327)	1868 (836)	6452 (11506)	4447 (8741)
	Stand	2724 (2429)	5068 (6796)	4043 (5332)	2621 (2674)	3767 (6432)	3265 (5027)
PNS Indicator (ms ² /Hz)	Rest	0.20 (0.14)	0.34 (0.16)	0.28 (0.16)	0.19 (0.14)	0.40 (0.24)	0.31 (0.22)
	Stand	0.06 (0.02)	0.12 (0.05)†	0.08 (0.04)*	0.06 (0.04)	0.12 (0.07)††	0.09 (0.06)***
SNS Indicator (ms ² /Hz)	Rest	3.39 (4.67)	1.64 (1.50)	2.40 (3.27)	1.78 (1.45)	1.24 (0.75)	1.47 (1.10)
	Stand	8.60 (4.54)	6.21 (2.91)	7.26 (3.77)*	8.70 (3.64)	5.76 (2.43)	7.05 (3.27)***

Legend: EC = Electronic cigarettes; HF = High frequency, LF = low frequency, PNS = parasympathetic nervous system, SNS = sympathetic nervous system, TC = tobacco cigarettes, * = $p < 0.05$ ("all" stand condition vs "all" rest condition - before smoking/vaping), *** = $p < 0.05$ ("all" stand condition after smoking/vaping vs "all" rest condition before smoking/vaping); † = $p < 0.05$ between group effect (TC vs EC before smoking/vaping, rest to stand transition); †† = $p < 0.05$ between group effect (TC vs EC after smoking/vaping, rest to stand transition).

TABLE 3 HRV responses of the participants.

can be inferred from the sympathetic indicator (a ratio of low frequency to high frequency power).

Our findings of similar resting circulatory and HRV measures between chronic TC-smokers and EC-users is supported by the work of Arastoo et al (10) who examined baseline, resting hemodynamics and HRV measures in 100 participants (42 chronic TC-smokers and 58 chronic EC-users). These authors found no difference between the groups on circulatory and HRV measures. They attributed these results to a consistent level of cardiac sympathetic activity within the two groups.

Secondly, this study demonstrated that circulatory parameters including HR, SBP and DBP as well as derivatives of these variables (MAP and RPP) were all significantly increased 10 min following the smoking as well as the vaping sessions. However, HRV measures were unchanged. Several studies compared the acute circulatory/hemodynamic responses in TC-smokers with EC-users. (8, 10, 16-20) One study also examined the acute effects of smoking/vaping on HRV. (10) Studies which have kept nicotine exposure equivalent between the groups, revealed that HR and/or BP are significantly increased to a greater extent in TC-smokers compared to EC-users. (8, 10, 16, 20) This augmented effect of TC-

smoking has been attributed to the greater number of chemicals (e.g., tar) in TC smoke. (10) These results indicate that TC-smoking may have a greater detrimental effect on the circulatory system and support the use of EC as a safer alternative to TC-smoking or as a smoking cessation strategy.

The findings of our study, however, agree with the findings of Franzen et al (17) and Vlachopoulos et al (19) whereby the increases in HR, SBP and DBP were comparable between the two groups. In these studies, the nicotine delivery may have been actually greater in the EC-use group leading to the equivalent increases in circulatory measures. Franzen et al (17) performed a randomized cross-over study with 15 active smokers. The physiological responses to TC smoking or EC-use (either with nicotine [24 mg/mL] or without nicotine) were examined in random order. HR, SBP, and DBP increased similarly in the nicotine conditions and but not in the EC condition without nicotine. The intensity of vaping was not standardized according to the depth of each breath during a puff. Thus, the amount of nicotine exposure could not be controlled. Vlachopoulos et al (19) also performed a randomized cross-over study with 24 TC smokers. Participants smoked either a TC or vaped

an EC without knowing the nicotine content. HR, SBP and DBP were monitored for 5-min as well as 30-min. Increases in HR, SBP and DBP were similar between smoking and vaping conditions at the 30-minute mark. Plasma nicotine levels were not assessed.

Both the tobacco cigarettes as well as the e-cigarettes used in this study contained nicotine. Nicotine stimulates the release of catecholamines and neurotransmitters and can cause tachycardia, and hypertension. (21) Nicotine also exerts pharmacologic effects which can enhance cardiac sympathetic activity. (5) Our finding of a negligible change in HRV dynamics in response to smoking/vaping is supported by the findings of Arastoo and colleagues. (10) These authors propose that the lack of change in HRV measures may be due to chronic tolerance of nicotine (by chronic smokers/vapers) and desensitization of central nicotinic receptors. Although sympathetic activity (as reflected by the SNS indicator) was not increased 10 minutes post-smoking/vaping, it is also possible that this parameter was increased during the smoking/vaping session and that this led to the increase observed in circulatory parameters. Moreover, withdrawal of PNS activity may also have contributed to the observed effects.

Finally, a unique aspect of this study was examining the circulatory and HRV response of smokers and vapers to an orthostatic challenge. Both groups responded similarly to the challenge, both before and after smoking/vaping. Postural change from the supine position to the standing position will induce a drop in BP due to venous pooling. (22) This drop in BP is detected by the baroreceptors which, in turn, will stimulate an increase in HR (via SNS activation and PNS withdrawal) and consequently will induce an increase in BP. This response was observed in this study and was not altered by smoking and vaping. It is also noteworthy that the orthostatic challenge placed a much greater physiological demand on the body than smoking/vaping.

4.1 | Limitations

This was a pilot study with a cross-sectional design and a small sample size ($n = 16$; TC = 7 and EC = 9). A

cross-over study could have increased the sample size and reduce the variability between the two groups. Secondly, a non-smoking comparison group was not included in the current study; a comparison group would have been useful to determine if the resting baseline circulatory HRV measures were increased due to an elevation in sympathetic activity. One study which examined resting HRV in healthy adults (aged 28 ± 8 years) indicated lower resting measures for HR (64 ± 10 bpm), SBP (108 ± 12 mmHg), DBP (64 ± 8 mmHg) and values for LF power ($734 [247-2389] \text{ ms}^2$) compared to the data obtained in our study. (23) It is also important to note that there are multiple factors which can influence changes in HRV such as; lifestyle, environmental, physiological, and neuropsychological factors. (24) These factors could have influenced our results. Also, we were unable to regulate the concentration of nicotine within the participant's cigarettes and vapes. In Canada, tobacco cigarettes contain between 8.0 – 18.3 mg (13.5 ± 0.49 mg) of tobacco which translates to 1.0 – 2.4 % ($1.80 \pm 0.06\%$) tobacco/cigarette. (25) The majority of EC users in this study vaped 5.0% nicotine (59 mg/ml) however two individuals used 3.0 % (39 mg/ml). (26) Ideally, blood samples to check for plasma nicotine concentrations following the smoke/vape session would have been obtained.

5 | CONCLUSIONS

The current study demonstrates that vaping has the same acute cardiovascular effects as smoking cigarettes in a small group of young adults. Thus, the use of e-cigarettes (vaping) containing nicotine may not be the safer alternative to cigarette smoking that is advertised. This discrepancy is significant as many individuals begin vaping because they believe it to be better for their health than cigarette smoking. With the increase in teens and young adults beginning to vape (27) more research on the cardiovascular response is needed to determine the relative safety and health risks associated with this behavior. It is important to educate the public regarding these risks, to make informed decisions re-

garding their health.

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The Development of the Evidence-Informed “OI Splint Kit” for Children with Osteogenesis Imperfecta and Their Families

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ABSTRACT

Background: Children with osteogenesis imperfecta (OI) live in fear of fracturing their bones. As fractures are unpredictable, there is a need for tools and knowledge to immobilize a fracture during emergencies. Inspired by a patient with OI recognized for fracture management, the aim of this patient-initiated project was to establish best practices for the safe splinting of fractures during emergencies, including the creation of an evidence-informed OI Splint Kit.

Methods: A clinical practice guideline study was conducted to create an evidence-informed splint kit for patients. A systematic review of the literature was conducted to identify kits and materials used to immobilize fractures with splints during emergencies. An expert Task Force was convened to review the results and a timeline was established to create a splint kit. The prototype underwent iterative cycles of modifications based on feedback from the Task Force.

Results: Four electronic databases were searched (Medline, CINAHL, PsychInfo, and Scopus), revealing zero publications pertaining to fracture immobilization kits. The Task Force used their clinical expertise and experiences to develop the OI Splint Kit. The following items were included: splinting tools, bilingual educational material, instructional cards, video tutorials, and memory card game.

Conclusion: A gap remains in terms of validated kits to assist in fracture immobilization during emergencies. A portable and practical OI Splint Kit was developed to assist in fracture immobilization emergencies, based on interprofessional clinical expertise and patient experiential knowledge. The kit is subject to ongoing testing quality evaluations to ensure its suitability for use in various contexts.

KEYWORDS

Osteogenesis imperfecta, Best practice guidelines, Splints, Fractures, Inter-professional care team

1 | INTRODUCTION

Affecting one in every 10,000 individuals, osteogenesis imperfecta (OI), also known as “brittle bone disease”, is a rare hereditary condition associated with bone fragility and fractures. (1) Consequently, children with OI live in fear of fracturing a bone which may curtail participation in activities and limit individuals who can handle them. (2-3) This extensive fear of fractures and situations that may cause fractures, ultimately affects children’s quality of life. (2, 4) In addition, the constant threat of any wrong movement or being mishandled also invokes fear of fractures in parents of children with OI. (2) As fractures may be inevitable and unplanned during the inherent ‘ups and downs’ associated with the daily lives of children with OI and their families, provisions are needed to optimally support them. (5-7) During these emergency situations, some children and their families have learned to temporarily immobilize their fractures to minimize the pain whilst being transported to medical facilities and/or awaiting treatment. Unfortunately, not all families are equipped with the knowledge, skills, and supplies to splint a fracture. (2) In the event of a fracture, readily accessible supplies and instructions may help immobilize the fracture, reduce the pain, help foster resilience, and offer some stress relief. (2) Establishing best practices with delineated safe handling procedures including the creation of an evidence-informed, portable splint kits offer a way forward to close a practice gap and help the OI community and those seeking to support them.

Throughout history, kits have proved to be useful and serve many purposes. The Indigenous people in the Plains region of Canada carried kits known as medicine bundles, which included a collection of spiritually significant items used for spiritual healing. (8) The widely known First Aid Kit, developed by the Johnson Johnson Family of Companies, was initially created to fulfill the need for readily available and accessible medical supplies to treat industrial injuries that occurred on railroad construction sites. (9) From there, the notion of a First Aid Kit, as a set of items needed to fulfill a specific purpose, has been shown to be clinically beneficial in many

areas of healthcare and in the community. In the pediatric clinical setting, Ballard and colleagues (10) developed two distraction kits containing items to help children manage procedural pain, and demonstrated that these kits were feasible and acceptable for use in pediatric emergency care. Kits created to treat medical emergencies, such as an opioid overdose, have been shown to be cost-effective and reduce opioid-overdose deaths. (11) Self-testing kits have also been developed, such as the Human Papillomavirus (HPV) kit used for cervical cancer screening. (12) These kits were convenient, easy to use, and cost-effective. Meanwhile, kits have also been created to facilitate knowledge translation. For instance, Freeman and colleagues (13) developed and evaluated the Youth KIT, a kit aimed at helping youth with disabilities during their transition into adulthood. The Youth KIT includes a set of modules and worksheets that focus on different aspects of adolescence, such as education and social activities, allowing youth with disabilities and others to better understand disability.

Hence, based on the collective benefits of other tested kits and inspired by the story of a patient and his family, clinical practice guidelines were developed to inform the creation of a portable splint kit for fracture management in OI. “...not if we break, but when we break.” (14) Carter Brown and his family know all too well the hardships following fractures. While learning to manage the multiple fractures to date, Carter and his mother, Jennifer Brown, have become resourceful in managing and immobilizing fractures during emergency situations. Their skillset was recognized in their local OI community. One afternoon, the Brown family was called upon by another family whose son with OI was discharged from the emergency room with an undiagnosed femur fracture. The fracture was undetected on x-ray; hence, the fracture and the accompanying severe pain went untreated. Still suffering, the other family did not know what to do and reached out to the Brown family for their expertise. The Brown family arrived with their ‘home grown’ splint kit to offer support. After the mother splinted the femur, instant pain relief was felt allowing the other family to re-group and seek medical care elsewhere. With this inherent reality associ-

ated with OI, Carter and his mother devised the idea of providing an “OI Splint Kit” to other OI families. They reached out to their hospital specialized in the treatment of OI for assistance. The Brown family sought to transform how children and their families manage fractures before arriving for emergency medical treatment, helping the global OI community. The family’s story of their commitment to help others made national headlines. Here lies how the Splint Kit was created in partnership with the family.

2 | METHODS

2.1 | Study Design, Goal, and Objectives

This clinical practice guideline study consisted of systematically reviewing the literature on existing kits or tools used to immobilize fractures during emergencies, as well as consulting key stakeholders, including patients, clinicians, and decision makers to create an evidence-informed portable “OI Splint Kit” for children with OI and their families. The methodology was guided by other evidence-based practice guidelines led by the research team and is described below. (6, 15)

2.2 | Expert Task Force Composition

An inter-professional Task Force was convened at the Shriners Hospitals for Children®-Canada (SHC-Canada), a university-affiliated, pediatric, orthopaedic hospital located in Montreal, Canada, and a member of the Brittle Bone Disorders Consortium. The 15-member Task Force was composed of healthcare professionals with expertise in OI, and included five registered nurses, 1 occupational therapist, 1 executive decision maker, 1 parent, 1 patient, 1 child life specialist, 1 pediatrician, 1 orthopaedic surgeon, and 1 scientist. Two undergraduate trainees from nursing and human anatomy were also part of the Task Force.

2.3 | Context and Setting

The development of the “OI Splint Kit” was conducted at the SHC-Canada where the OI program was founded in the 1990s and became internationally acclaimed as the standard of OI care. (16) The hospital provides care to a global cohort of children with OI. In 2014, it created and implemented the *Family and Patient Focused Care, Open Communication, Collaborative, Understanding and Compassionate, Safe and Seamless, Expertise and Education, and Driven by Research and Best Practices* (FOCUS^{ED}[™]) philosophy of care, which guides the practice of all hospital staff. (17) The creation of the OI Splint Kit aligned with this philosophy of care.

2.4 | Literature Review, Extraction, Appraisal, and Synthesis

The search strategies were developed in collaboration with a librarian scientist, which consisted of subject headings and/or keywords relevant to “splint kits”, “fracture immobilization”, “osteogenesis imperfecta”, and “emergencies”. The following four electronic databases were searched for articles that suited the inclusion criteria (**Table 1**): Medline via Ovid (1980 to 2020), CINAHL via EBSCO-host (1980-2020), PsychInfo (2000-2020), and Scopus (1980-2020). Any discrepancies that arose in the process of the literature search were resolved by discussion with members of the Task Force until consensus was achieved. These discussions continued throughout the study duration.

2.5 | Consensus Development Based on Evidence

The Task Force reviewed the search strategy and results, prioritized relevant key items required for fracture immobilization, and established a timeline. The impetus for Splint Kit began in 2017 and was finalized in 2019. Iterative circulation of suggested items permitted the Task Force to determine if proposed content: (a) was suited for children with OI during fracture emergencies; and (b) could be understood by children and their fam-

ily caregivers during emergencies. Items included materials and instructional methods (cue cards and videos) needed to immobilize a fracture and led to the creation of a game to help children learn about their anatomy and provide distraction. Drafts of the OI Splint Kit prototype were circulated in person and by email to elicit feedback, obtain constructive comments, and establish consensus. The Task Force subsequently contributed to presenting the OI Splint Kit at an international OI meeting and drafted the manuscript.

2.6 | Guidelines and Conflict of Interest

The Task Force members requested that other clinicians and the OI community (e.g. individuals with OI and their families) provide critical, written and/or verbal feedback on the content, clarity, and utility of the Splint Kit. The kit was confirmed to be comprehensive and easy to use, with contents appropriate for splinting a child with OI following a fracture. The Task Force members did not have any conflicts of interest that may have altered the professional feedback and consensus generation. Furthermore, the study was not related to any financial or commercial activity.

2.7 | Revisions Dates and Plans for Updates

The development of the OI Splint Kit was in accordance with the current, clinically adopted ways of immobilizing children with OI at SHC-Canada. The OI Splint Kit responded to the idea of a child and his family desiring that all children have the knowledge and skills to be immediately immobilized following a fracture. (14) Since the creation of the Splint Kit, over 300 kits were distributed to children with OI and their families by their nurse coordinator inviting opportunities for teaching by members of the inter-professional team. Subsequently, a process was set-up at the hospital for families to purchase additional kits and replenish their supplies using the hospital's website. Ongoing fundraising campaigns by Carter and his family ensued to provide additional kits for school. Like any tool, guideline, or resource, the creation of the OI Splint Kit was a dynamic process requiring feedback from the inter-professional team including children with OI and their families. Therefore, the OI Splint Kit is subject to continuous review to remain clinically relevant, meaningful, and integrated into practice. Quality improvement evaluations are conducted to: (a) assess the usefulness of the kit during emergency fractures; (b) gauge convenience (e.g. daily portability); and

Search Strategy for Medline via Ovid (1980-2020), PsychInfo (2000-2020), and SCOPUS (1980-2020)		
Search	Keywords	Results
1	Osteogenesis Imperfecta OR OI OR Brittle Bone Disease	27,336
2	Splints/External Fixators/Medical Therapeutic Devices OR Splinting Tools OR Splint Kit	38,404
3	Emergency Management/Preparedness OR ER	832,840
4	S1 AND S2 AND S3	0
Selection Criteria		
	Inclusion Criteria	Exclusion Criteria
Population: Age and Diagnosis	All ages with diagnosis of any type of Osteogenesis Imperfecta	No diagnosis of any type of Osteogenesis Imperfecta
Study Design	Qualitative, quantitative, and mixed methods, abstracts, review articles, commentaries	None
Language	English or French	Any other language

TABLE 1 Search Strategy and Selection Criteria

(c) determine if items require adaptation, addition, or removal. The Task Force will reconvene yearly to discuss revisions and modifications. We invite the OI community to adapt and implement these guidelines in their varying settings and contexts to help improve the safe handling of children with OI following a fracture.

3 | RESULTS

3.1 | Search Results

Four literature database searches revealed zero publications pertaining to the subject headings and/or keywords (Table 1). As the electronic searches revealed zero results, no titles, abstracts, or full-text articles were screened or reviewed. The last search attempt occurred on February 28, 2020 following the creation of the Splint Kit to re-confirm the zero search result.

3.2 | Compilation of Clinical Evidence

Due to the paucity of literature available for the creation of the Splint Kit, the Task Force used their clinical expertise and patient experiences to prioritize a list of items for inclusion (Table 2). Some of these items may be purchased at a local pharmacy (items 4 to 15 in Table 2); however, other items were synthesized by members of the Task Force, including the bilingual educational material, available in print, video and digital formats, to support the individual immobilizing the child following a fracture (items 1 to 3 and 16 to 23 in Table 2). The main immobilization procedures were illustrated on individual cards and served as a reminder of the main steps used for immobilization (Figure 1). Fourteen videos were created in French and English (83 minute and 43 seconds [English: 42 minutes and 35 seconds; French: 41 minutes and 8 seconds]) offering detailed instructions on how to immobilize a child (Figure 1) and ideally watched before a fracture occurs. These videos are available for viewing on the Vimeo website and children and parents were encouraged to share them with the child's network (e.g. grandparents, daycares, and schools) (Table 2).

Finally, a trilingual game (in French, Spanish and En-

glish) was created for the children called "Bones and Fractures Memory Game" (Figure 2), and is available as Supplemental Material. A preliminary list of terms and one-sentence descriptors were composed and validated by the Task Force. The nurses, pediatrician and cast technicians were instrumental in validating the common fractures and treatments associated with OI. They also selected terms that are used in everyday practice. The child life specialists aided in the one-sentence descriptors ensuring that the vocabulary was age-appropriate and clinically relevant. Changes were made to reflect their feedback. This iterative process occurred over three different cycles with the Task Force. The final prototype comprised 26 cards distributed over types of bones, fractures, and treatment options. The card images were subsequently illustrated by the study site graphic designer and validated by the clinicians to ensure the images accurately depicted the terms. A questionnaire (Appendix I) was created to evaluate various aspects of the game from the child's perspective, which was filled out by the children after one round of play with a member of the Task Force. Based on their comments, modifications were made to the game and the prototype was finalized.

4 | DISCUSSION

Our literature search revealed that no splint kits were published in the OI literature despite fractures serving as a primary feature of OI and source of fear for children and their families. (2, 7) In an effort to quickly address this gap in the quality of care and health services, our inter-professional team opted to translate their knowledge into a tangible, practical solution in partnership with Carter Brown and his family. The iterative process of creating the OI Splint Kit relied on the collective experience of Task Force team members who practice in an internationally-renowned hospital for the provision of OI care. Ongoing quality improvement evaluations will permit the OI team to revise and modify the OI Splint Kit as needed and continue to solicit input and offer their expertise to the global OI community.



FIGURE 1 Instructional Methods Used to Teach Children and Family Caregivers How to Splint a Child with OI following a Fracture.

Carter Brown showcasing the contents of the splint kit. Instructional videos created by the cast room nurses, publicly available on Vimeo for viewing on Vimeo at <https://vimeo.com/showcase/5149282>.

When a fracture is suspected, children with OI and families are advised to immediately splint the fracture to minimize the acute pain and post-structural deformities, which may lessen the risk of developing chronic pain. (18-20) However, a fracture may occur *anywhere* or *anytime* and not necessarily with adequate skills, supplies, and personnel to treat it. During this emergency, children, their families, and other responders (e.g. teachers or lay person), often need to creatively problem-solve to successfully immobilize the fracture, decide *if*, *how*, and *where* to seek emergency help for their rare condition. Further, individuals with OI must remain vigilant of further injury from clinicians who “Don’t know how to treat [them]!” (21) The OI Splint Kit serves as a tool to bridge the gap between unprecedented fracture incidents and treatment accessibility including the provision

of instructions which may be shared with clinicians unfamiliar with OI.

We further encourage the use of the OI Splint Kit with the completion of the OI Good to Go Passport and other passports created by the OI Foundation (OIF) and OI Foundation of Europe (OIFE) to further communicate this rare condition to responders. (6) The OI Good to Go Passport is a wallet-sized card containing information to accommodate the transitioning needs of the OI population, including their medical condition and diagnosis, surgical history, medications, and contact information of their specific OI healthcare team. (6) Moreover, we encourage the adoption of the OI Splint Kit to foster children’s participation in their care (22) and reinforce collective efforts to prepare the transition of the OI population into adult-oriented health care systems. (6, 15,

Item	Description and Rationale	Unit #
1. Instructional Card #1	Illustration card: brief rationale of the project and links to the videos.	1
2. Instructional Card #2	Illustration of the main immobilization for upper extremities fracture (retro-verso: arm and forearm).	1
3. Instructional Card #3	Illustration of the main immobilization for lower extremities fracture (retro-verso: upper and lower leg).	1
4. ACE Bandage with Self Closure (3in)	An elastic bandage that may be wrapped around an injured area to control swelling or around a cast to secure it in place.	1
5. ACE Bandage with Self Closure (4in)	An elastic bandage that may be wrapped around an injured area to control swelling or around a cast to secure it in place.	1
6. 3M Coban Cohesive Bandage (3in)	A self-adherent elastic tape that may be wrapped around an injured area to control swelling or around a cast to secure it in place.	1
7. 3M Coban Cohesive Bandage (4in)	A self-adherent elastic tape that may be wrapped around an injured area to control swelling or around a cast to secure it in place.	1
8. Padding Cast (7.5cm)	Cotton bandage that may be wrapped around an injured area to provide padding and prevent friction.	1
9. Padding Cast (10cm)	Cotton bandage that may be wrapped around an injured area to provide padding and prevent friction.	1
10. One-Step Splint (3x35in)	A splint made of two layers of padding covering a fiberglass slab for an easy one step immobilization.	1
11. Scissors (5.5in)	To help cut the cast.	1
12. Stockinette	A thin layer of fabric that is applied directly on the skin when casting, preventing movement of the cotton padding and friction from the cast edge.	1
13. Human Bone Stress Ball	To help relieve stress and muscle tension. May be used as a distraction tool.	1
14. Drawstring bag	A bag to carry all the tools needed to immobilize a fracture.	1
15. Sam Splint (5.5x36in)	Splint made of soft aluminum with a foam coating that is applied to an injured area to stabilize a fracture.	1
16. Video #9	<i>Trousse de premiers soins OI - Immobilisation de la cheville ou du pied.</i>	1
17. Video #10	<i>Trousse de premiers soins OI - Immobilisation du bras.</i>	1
18. Video #11	<i>Trousse de premiers soins OI - Immobilisation de l'avant bras.</i>	1
19. Video #12	<i>OI Support Kit - Immobilization of the Forearm.</i>	1
20. Video #13	<i>OI Support Kit - Immobilization of the Femur</i>	1
21. Video #14	<i>OI Support Kit - Immobilization of the Arm.</i>	1
22. USB Key	Contains the content of the instructional cards and videos.	1
23. Bones and Fractures Memory Game	Helps children learn about their bones, fractures, and different therapies used in OI. Offers a potential distraction tool.	1

TABLE 2 Description of Contents Contained in the Splint Kit Created for Children with Osteogenesis Imperfecta Following a Fracture and Requiring Immobilization

21) However, if and how to integrate the OI Splint Kit into the OI programs of various hospitals warrants discussion, decisions, and action at a local level, and may be supported by local OI foundations and other key stakeholders as well.

At every point of care, patients have unique experiences that make them knowledgeable about their illness and the care they receive. Thus, healthcare professionals need to create collaborative partnerships with patients and their families to optimize the provision of care.

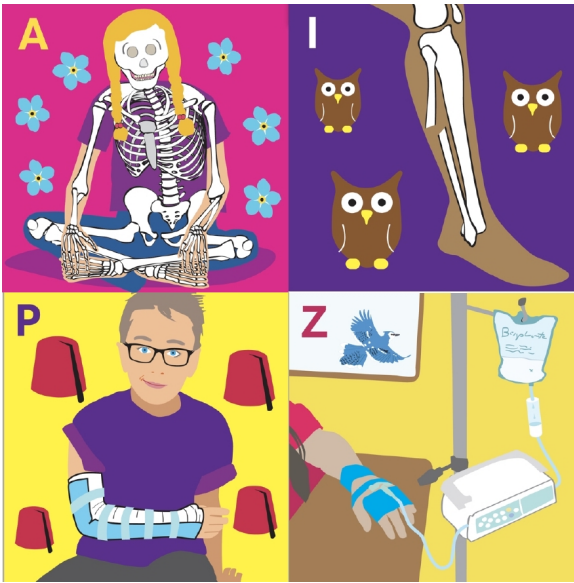


FIGURE 2 Examples of the Cards included the Bones and Fractures Memory Game

There are 26 cards (A to Z) included the Bones and Fractures Memory Game from. A = Human Skeleton (206 bones); I = Displaced Fractures; P = Backslab; Z = Bisphosphonate. The game is available in English, French, and Spanish.

“Patients as Partner” is a local program that encourages patients to express their opinions regarding their health and illness as a full member of the clinical team. This innovative and viable approach recognizes the experiential knowledge of patients allowing for the creative adaptation of practices to improve quality of life and establish more meaningful interactions with professionals. (23-24) Our approach, guided by our FOCUSED™ philosophy of care (16), recognized the lived experiences of Carter Brown’s and family and their expertise in adapting to unexpected situations. Similarly, Pomey and colleagues (23-24) concurred that chronically ill patients and their caregivers develop extensive knowledge concerning their symptoms and the effects of different interventions on their state of health and on their lives. Thus, creating partnerships with patients and their families to help inter-professional health care teams and researchers create innovations to help with their health care is critical.

4.1 | Strengths and Limitations

Without any published empirical knowledge, the development of the OI Splint Kit drew on the experiential knowledge of OI experts in partnership with a family with OI. Efforts to remove potential biases associated with the development of the OI Splint Kit included the formation of an inter-professional task force, acknowledgment of no conflicts of interests, and the adoption of a rigorous methodology to critically appraise and synthesize all available evidence. (6, 15) The OI Splint Kit was created in collaboration with a leading institution in the provision of OI and generated significant praises by the cohort of children and their families followed at the study site and at an international OI meeting. Yet, the OI Splint Kit may not be generalizable or transferable to all contexts, and there is no research to confirm its effectiveness. As the OI Splint Kit is meant for emergencies, the kit focuses on splinting methods and tools in such cases. Splinting is a safe and easy-to-replicate method for non-healthcare professionals in a home setting and reduces the risk of leverage fractures in the OI population. Thus, other methods, such as the use of plaster to immobilize fractures were not considered appropriate by our Task Force for use in the splint kit, or in our literature review. Similar to other guideline creations, clinical reasoning is essential for the clinicians to ensure best clinical care, especially since the OI Splint Kit has not been evaluated in practice. Furthermore, these kits need to be adapted to local contexts, expertise and resources.

4.2 | Future Directions

While other kits used in the pediatric healthcare setting have shown clinical efficacy (10-13), the OI Splint Kit requires a rigorous evaluation to determine its benefits in fracture management. To date, the OI Splint Kit has received informal feedback from patients and health care professionals, supporting its benefits. Albeit, evidence for the usefulness and effectiveness of the kit remains anecdotal. We will start a quality improvement initiative, guided by a leading implementation science framework

(25), to evaluate the OI Splint Kit. Namely, our future testing will give critical insight on the following points: (i) did the kit aid in fracture immobilization during emergencies; (ii) frequency of usage of the kit; (iii) did the kit help decrease pain; (iv) did the kit help ease anxiety during fractures; and (v) is the OI community satisfied with the kit. The evaluation will also collect requests for modifications to the OI Splint Kit. The comments will be shared with our inter-professional Task Force, and together with the research team, the kit will be iterated to satisfy the needs of the OI community. Implementation efforts will ensue to make the OI Splint Kit available in the long-term to the global OI population.

5 | CONCLUSION

Children with OI and their families live with the constant risk of unexpected fractures associated with their genetic condition creating distress and potentially diminishing one's quality of life. (2) The creation of the OI Splint Kit was in response to one family who desired to share their practical solution to cope with fears of fractures with other families. (2) Driven by a FOCUSED™ philosophy of care (17), and the multitude of benefits of partnering with children, families and the inter-professional team, an evidence-informed kit was created to help children and their families be better prepared during an emergency. While we encourage the adoption of the OI Splint Kit into practice, we remain mindful the kit has not been subjected to rigorous testing and rely on the clinical reasoning of the clinicians to determine if this kit is suitable for use in their local context and properly adapted to local expertise and resources.

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erosity of many donors permitted the provision of a free splint kit to each of the 360 families living with OI at Shriners Hospitals for Children-Canada.

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Understanding Women's Anxiety and Uncertainty Attending a Rapid Diagnostic Clinic for Suspicious Breast Abnormality: A Mixed Methods Study

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ABSTRACT

Background: Rapid diagnostic centres (RDC) for breast abnormality offer a speedier process from the discovery of a suspicious breast lump to same-day investigation and confirmation of a breast cancer diagnosis.

Purpose: This proof of concept study aimed to assess the anxiety and uncertainty levels of women going through an RDC and explore women's need for support during the diagnostic period.

Methods: Thirteen women who attended an RDC in 2013 took part in a sequential mixed-method study to assess anxiety and uncertainty levels. Measures were taken pre-and post-testing, at three weeks following receipt of results, and were followed by a semi-structured telephone interview.

Results: The mixed data results show congruence between women scoring above clinical values for anxiety and above normative values for uncertainty and detailing their RDC experience as stressful. At pre-diagnosis, uncertainty and anxiety levels were above clinical and normative values for the majority of the thirteen women. Among the women who received a cancer diagnosis (7/13), five had high anxiety, and two scored above normative values for uncertainty. Among the women with a benign diagnosis (6/13), all had anxiety scores below clinical levels, and three had scores above normative values for uncertainty. Anxiety and uncertainty levels remained relatively the same from the three days to three weeks post-testing. The women suggested the need to receive details of the day's unfolding, especially what medical procedures will take place, how, and why, and in advance of the day of testing.

Conclusion: While RDCs offer women with a suspicious breast lump the opportunity for quicker diagnostic testing, preliminary results suggest that the period leading up to the day of testing and three days and three weeks post-testing is marked with anxiety and uncertainty levels above clinical and normative values. The results illustrate the need for further inquiry into the psychological impact of obtaining testing at RDCs for a breast abnormality. Results suggest a potential role for nurses to support the waiting period with psycho-educational guidance and resources.

KEYWORDS

rapid diagnostic centre, breast cancer, psycho-educational support, uncertainty, anxiety

1 | INTRODUCTION

During the diagnostic period after a breast lump is discovered or a mammogram displays suspicious findings, women experience significant psychological distress, including anxiety, uncertainty, and symptoms of acute stress reaction. (1-5) A systematic review of thirty research studies with large sample sizes examining the presence of anxiety levels in the diagnostic period for women suspected of breast cancer found that, on average, 8% to 50% experienced high levels of anxiety. (2) The high anxiety was also found to persist throughout the diagnostic evaluation until certitude was achieved by establishing the definitive diagnosis. (6) Such high levels of anxiety can easily disrupt an individual's ability to maintain their everyday activities and are also documented as having a negative effect on the immune system. (7-9) Pre-diagnostic anxiety is a significant predictor of post-diagnostic anxiety that can impact a person's ability to cope and have implications for adverse long-term outcomes. (2,10) Although the diagnostic phase can be marked with intense emotional distress, this phase is often overlooked in research. (3,11) Rapid diagnostic centres (RDCs) or one-stop clinics were developed to improve the diagnostic process for women and offer same-day investigation and a quick turnaround for a diagnosis of breast cancer (same day to three days post-investigation). (3,12) Conceptually, the idea of RDCs for women with suspicious breast abnormalities is attractive, especially to reduce wait times to diagnosis. (13) However, for the few studies that have investigated the psychological impact of rapid diagnostic testing for breast cancer, the consensus remains unclear regarding its effects on the individual's anxiety, uncertainty, and stress (3,4,14-16); nonetheless, current longitudinal evidence is pointing towards a positive association between symptomatic breast disease and psychological distress. (2,4,17) In addition, there is limited information on whether receiving psychological support during the rapid diagnostic process is needed and valuable, and if so, in what forms. (18) The objectives of this proof of concept study were to assess these outstanding gaps by measuring and describing the levels of

anxiety and uncertainty as experienced by women undergoing rapid diagnostic testing for a suspicious breast abnormality before and after testing and exploring the women-expressed needs for support during the diagnostic period.

2 | THEORETICAL MODEL

The Mishel Uncertainty in Illness model guided this study. (19) Mishel ((19), p. 225) defines uncertainty as "the inability to determine the meaning of illness-related events." Uncertainty arises when a person cannot characterize an event because of insufficient cues. This inability may be especially acute in a complex situation, such as a diagnostic workup, where the patient is overwhelmed with an abundance of unfamiliar cues. When a situation is appraised negatively, uncertainty is viewed as a threat rather than an opportunity. In these instances, uncertainty becomes a covariate of anxiety. (20) Thus, one can anticipate that a reduction in uncertainty could, in turn, lead to a reduction in anxiety. (4,21)

3 | METHODS

The setting for this preliminary proof of concept study occurred in an RDC situated in a large Canadian cancer research, treatment, and educational centre. The study followed a sequential mixed-method design. A quantitative phase was first used, followed by a qualitative phase to describe and understand the anxiety and uncertainty experienced by women undergoing rapid diagnostic testing for a suspicious breast abnormality. The quantitative data were collected at three-time points [at pre-diagnosis (T1), three days post-diagnosis (T2), and three weeks post-diagnosis (T3)]. Using the interview format, we collected the women's accounts of their experience at three weeks post-testing, coinciding with quantitative data's last time point measurement. The purpose was to deepen the understanding of the women's scores on the anxiety and uncertainty scales as experienced during the rapid diagnostic phase and obtain their views on the type of support needed during the diagnostic

phase. Data collection occurred in 2013. Research ethics approval was obtained from the Research Ethics Board of the participating hospital 14-092-CE.

3.1 | Sampling

Inclusion criteria included women from the hospital's catchment area who had a suspicious breast abnormality that was detected radiologically or clinically, with no previous history of breast cancer diagnosis, who had an appointment at the RDC and did not have a known history of *BRCA1/BRCA2* mutations. Women were excluded if they did not read or speak English, were less than 18 years of age, had a recent fine needle aspiration that was suspicious for malignancy, or had pre-existing anxiety or major depressive disorder diagnosis. A convenient sampling approach was used to select women. Women who met the inclusion criteria were identified and approached initially by the clinic's medical administrative assistant using an information script. If the women were interested in participating, they were asked for verbal permission to forward their name and telephone number to the study research assistant, who arranged for and conducted the informed consent process. Only those that provided written consent were included in the analysis. Twenty-four women were approached for the study. In total, 13 women met the eligibility criteria and agreed to participate. Reasons for declining were not interested in the study and could not commit time to conduct the interview.

3.2 | Data Collection and Measures

The quantitative data collected included demographics and two validated scales measuring uncertainty and anxiety. Uncertainty was measured using Mishel Uncertainty in Illness Scale - Community form (MUIS-C). (22,23) The MUIS-C is a 23-item scale with responses rated on a 5-point Likert scale ranging from 23 to 115 with a mid-range score of 69 and normative values for breast cancer identified at 33.7. (22-24) In our study, alpha coefficients ranged from 0.88 - 0.97. Anxiety was measured using Spielberger's State-Anxiety scale (STAI-

S), (25,26) a 20-item scale with responses rated on a 4-point Likert scale ranging from 20 - 80. The normally recognized score of 40 for the clinically significant value of symptoms of a state of anxiety was used in this study. (27,28) Alpha coefficients in our study ranged from 0.94 - 0.97. Questionnaires were administered over the telephone; the participant had a copy of the questionnaire to follow along while the research assistant read out the questions and answers. The questionnaires were administered at pre-diagnosis (T1), three days post-diagnosis (T2), and three weeks post-diagnosis (T3). At T3, the principal investigator contacted the women to invite them to participate in a telephone semi-structured interview that focused on 1) the participant's perception of the diagnostic process, 2) challenges faced during this process, 3) their views on areas of uncertainty and anxiety during this process, and 4) suggestions for service improvement. Examples of questions from the interview included: "Tell me what it was like waiting for further testing and waiting for your results? What feelings or concerns did you have during the waiting times? Was waiting for your test results stressful for you? What was it like "not knowing"? What suggestions would you have to improve the diagnostic process?" All participants were interviewed except one who was lost to follow-up following T2. All interviews were audio-recorded and transcribed verbatim.

3.3 | Data Analysis

Quantitative data: The statistical software SPSS 20 was used to generate percentages, means, standard deviations, and reliability scores (Cronbach's alpha), as well as non-parametric statistical analyses to test for significance and correlation from the quantitative data generated from the study survey: MUIS-C (uncertainty outcome) and STAI-S (anxiety outcome). Descriptive statistics were used to summarize the participants' characteristics and levels of uncertainty and anxiety. Pearson correlation coefficient was used to test the association between anxiety and uncertainty levels.

Qualitative data: Qualitative data generated from the semi-structured interviews were transcribed verbatim

and coded using content analysis (29,30) to develop the categories and themes that represented the aggregate data. Three members (CM, CW, DH) independently coded the data. Interview coding continued until a consensus of coding categories and thematic saturation was reached.

Triangulation of Quantitative and Qualitative Data: This study followed a mixed, quantitative-qualitative, sequential analytical approach in which the quantitative data was dominant and analyzed first, followed by the analysis of the qualitative data, used as an adjunct to understand the quantitative findings further. (31) A matrix was developed to show high and low scores of anxiety and uncertainty, with the qualitative data providing a deeper understanding of the quantitative emotional values observed during and after testing. The same three investigators independently reviewed the integration of quantitative and qualitative data and then met as a team to discuss the final findings until consensus was reached.

4 | RESULTS

The thirteen women who took part in the study are representative of the general population seen at the RDC. They had a mean age of 50, had an average of two children, the majority were married (n=8/13) and university-educated (n=8/13). While about 15%-20% of all breast cancer cases tend to be familial (32), in this sample, there was a higher sample of women with a family history of the diseases (n=7/13). Following testing, among the 13 women, seven were found with a malignant tumour and six with a benign tumour.

At the pre-diagnosis, nine out of thirteen women scored above the clinical values for symptoms of state anxiety and all thirteen scored above normative values for the state of uncertainty (normative mean value = 33.7, standard deviation + 12.9; a range of 23–115) (see **Table 1**). A two-tailed Pearson correlation coefficient test between anxiety and uncertainty revealed a strong association of 0.757 ($p=0.003$).

The following categories separate the triangulated re-

sults from the quantitative and qualitative data to reveal themes that could help explain their emotional experience: 1) by the time of diagnosis from pre-diagnosis to three days and three weeks post-diagnosis and 2) by below and above clinical and normative values for anxiety and uncertainty.

4.1 | Pre-diagnosis: Below clinical values on anxiety but above for uncertainty

Two themes best describe women's experiences with low anxiety (n = 4/13) but high uncertainty (13/13): use of positive reinterpretation and support received by clinical staff.

Use of Positive Reinterpretation: The four women in this category, although experiencing high levels of uncertainty, maintained their anxiety level below the cut-off by viewing their situation in a more favourable light, such as an opportunity. That is, whenever they worried about their upcoming test results, the women described using repetitive positive thoughts and holding on to the belief that their suspicious breast abnormality would probably be benign. They viewed their situation as positive: "it is just the healthcare team erring on the side of caution" (Participant 2). The four women also described the use of calming self-talk to help reinterpret negative anxiety-provoking thoughts into positive thoughts "I am able to get through it. I can deal with this... I am strong" (Participant 1). Another woman described how she practiced letting go of a situation she had no control over and how that process left her feeling less anxious "so I said to myself, until I have more information, I am just going to leave it and not try to think about what it could be" (Participant 2).

Support by clinical team: All the four women in this category mentioned how the support and reassurance they received from the clinical team helped them remain calm: "it is most likely not cancer" (Participant 9). The clinical nurses' explanations as to why further tests were needed were mentioned as helpful to reduce uncertainty and anxiety.

Timepoint	Anxiety Above Cut-Off (40)	Uncertainty Above Normative Values BC (33.7)
T1 Pre-Diagnosis	Malignant n=3 Benign n=6 Total n=9	Malignant n=6 Benign n=7 Total n=13
T2 3 Days Post-Diagnosis	Malignant n=5 Benign n=0 Total n=5	Malignant n=2 Benign n=3 Total n=6
T3 3 Weeks Post-Diagnosis	Malignant n=3 Benign n=2 Total n=5	Malignant n=4 Benign n=3 Total n=7

TABLE 1 Uncertainty and Anxiety above clinical cut-off by diagnosis received. Malignant, n=7, Benign, n = 6, Total, n=13.

4.2 | Pre-diagnosis: Above clinical values for both anxiety and uncertainty

For both groups of women with a malignant and benign diagnosis whose scores were above clinical values at pre-diagnosis for anxiety (n = 9/13) and uncertainty (n = 13/13), two themes best describe their experiences: 1) additional testing generating uncertainty, and 2) concern over maintaining responsibilities.

Additional testing generating uncertainty: These women described a lack of understanding of the additional testing being carried out and how additional testing led to considerable uncertainty. They also talked about feeling anxious while waiting for the test results from their additional testing. One woman described her anxiety arising from a statement made by her health professional that the participant could not decipher the true meaning of “they want to do a biopsy to check on some cells... What exactly did she mean? I consider this a statement with no ending” (Participant 8). To another woman, the feeling of uncertainty occurred because the necessity of additional tests made it hard for her to view the situation in any other way than “catastrophic” (Participant 9). Notwithstanding the small sample of nine, the findings above are aligned with the association found between the quantitative measures of anxiety and uncertainty before receiving their diagnostic results ($p=0.003$).

Concerns over maintaining responsibilities: Women who

feared the "worst-case scenario.": Women who expressed fears and increased perception of being found with cancer described how they were affected by intrusive thoughts of cancer and had difficulty carrying out their daily responsibilities such as childcare. They described their pre-emptive worrying about the potential cancer diagnosis and recounted their thoughts about how it could negatively and significantly affect their obligations, responsibilities, and lifestyle, citing examples such as their ability to be a good mother. For instance, a mother with a young child at home described the situation in this way:

“... I was worried; I have a seven-year-old boy, and when you do not know... what is happening, you think about the worst-case scenario...cancer and then the treatment and how this is going to affect your lifestyle and the fact that you have a...very young child. I worry about the future as a mother”

Participant 5

In expecting the worst-case scenario, these women explained their experience prior to testing as being frozen by their fear and having difficulty functioning in their daily tasks. Finally, one contextual similarity was found among the women of this category who exceeded clinical and normative values for both anxiety and uncer-

tainty: their shared family history with cancer. They experienced the highest anxiety levels, with scores up to 73 with a possible maximum score of 80. (25,26)

4.3 | Three days post-diagnosis: Above clinical values for anxiety and uncertainty among the women who received a cancer diagnosis

Among the women who received a cancer diagnosis (7/13), five had high anxiety, and two scored above normative values for uncertainty. Three major themes were described by those receiving a cancer diagnosis: 1) as "life-changing"; 2) as having to face many uncertainties and stressors, and 3) for two women, "a relief."

Cancer is life-changing: These women described a cancer diagnosis as life-changing with many uncertainties. One of the women captured this feeling when she explained how she felt after hearing she has breast cancer, "I mean, how can you not be upset? Your whole life is completely discombobulated at that point because you have so many things to think about, and your whole life is going to completely change" (Participant 3). The women described feeling anxious about how they would maintain their daily routines for themselves and others, such as their weekly physical activity routine. Half (4/7) of the women described how overwhelmed they felt and how upset they were at their bodies for having let them down. They portrayed the situation as one of the most stressful events of their lives. However, some women said that they felt thankful for the way the healthcare team attempted to reassure them, providing them with immediate details of treatment plans and that they felt supported as they moved into the next phase of being a cancer patient. In the days following the diagnosis, the women described how they reached out for support from family, with one specifying how she sought refuge in her religious faith.

Facing uncertainties and stressors: The uncertainty of their treatment plan and the unfamiliarity with potential side-effects left women feeling anxious: "...I have heard that you are really sick in chemo, but are you sick the whole time or are you just sick on certain days

and then the whole idea of losing your hair" (Participant 3). Another woman described how her treatment plan, which consisted of chemotherapy, was going to affect her ability to enjoy the seasons and holidays: "...I knew it was a year, a year by the time you go through all this (chemotherapy). So then in your mind, you are thinking, okay, well I am going to miss...the fall and Christmas. This is not going to be great" (Participant 8). Women went through additional testing such as a biopsy to obtain their diagnosis. One woman described this experience as a "funeral moment".

A sense of relief: The sense of relief expressed by two women in the cancer group was described by three certainties: "know[ing] what the [diagnosis] was" (Participant 3), "knowing cancer had not metastasized" (Participant 8), and "being recommended treatment that did not include mastectomy" (Participant 11). Knowing their breast abnormality was cancer provided a sense of certainty to some women. They now knew what they were dealing with (cancer) and could formulate treatment plans in line with their wishes. This, in turn, gave them a feeling of control over the situation. As described by one of the women, "When they recommended a lumpectomy...tsunami of relief rush[ed] through me because I was so afraid of losing my breast that when they said lumpectomy, I was like, fine...where do I sign?" (Participant 11).

4.4 | Three days post-diagnosis: Above clinical values for anxiety and uncertainty among the women who received a benign diagnosis

Among the women who received a benign diagnosis (n = 6/13), all had anxiety scores below clinical levels, and three had scores above normative values for uncertainty. Two themes describe their experience post-diagnosis for this subgroup: a) absolute relief and b) adjusting to the diagnosis.

Absolute relief: The six women described a sense of relief upon hearing that their suspicious breast lump was benign. The women described the event as a huge weight taken off their shoulders, allowing them to return

to their daily routine and habits, as well as validating their initial instincts that the lump would be found to be benign or their suspicions unfounded. One woman (Participant 10) who had advocated for a referral to the RDC to receive additional testing to confirm a previous diagnosis of benign fibroadenoma explained that she felt relief from having it "confirmed" by a biopsy.

Adjusting to the diagnosis: For the three women who scored above normative values for uncertainty three days post-diagnosis, their uncertainty levels remained above three weeks post-diagnosis. However, two of these three women who received a benign diagnosis were told they would need a lumpectomy and further pathological testing to remove the abnormal tissue formation. They described the need for further investigation using language that was suggestive of residual uncertainty, such as how tumour growth is unpredictable, no diagnosis is ever 100% certain, that this is why they want to remove the abnormal growth, and we cannot predict every single detail of what is going to happen next. Specifically, the women used words such as "So...when I got the diagnosis that it is a great possibility that it is 100% benign, but there is still a chance..." (Participant 4) and "there is a possibility that it could be a type of benign tumour...that could get larger, so the recommendation is to have it removed" (Participant 7). Another woman who presented with residual uncertainty three days post-diagnosis despite receiving a benign diagnosis reported that she understood her diagnosis as having both a malignant and benign form of cancer that could later develop into full cancer.

4.5 | Three weeks post-diagnosis: Above clinical values for anxiety and uncertainty

Anxiety and uncertainty levels remained relatively the same for both groups of women with a malignant and benign diagnosis from three days to three weeks post-testing. There was, however, an increase noted in the benign group, with two now experiencing anxiety above the clinical levels and an additional woman experiencing uncertainty above normative values. The two women

who now experienced high anxiety at three weeks post-testing described that they expected to receive a cancer diagnosis eventually. One woman described her belief as "the negative now is that I am waiting for it...I missed this one...but I figure it will hit me at 60, 65...so you know other women would say, oh my God, it is a blessing, it is fantastic, I am saved! No, I am waiting. I am waiting for the shoe to drop" (Participant 5).

4.6 | The need for support during the diagnostic period

The women suggested the need to receive details of the day's unfolding, especially what medical procedures will take place, how, and why, and in advance of the day of testing. They wanted to know if they needed to take pain medication prior to their clinic appointment for further testing and if it was best to be accompanied. On the day of receiving their results, half of the women shared needing more information on their diagnosis, such as a written report on the stage of their diagnosis. One suggestion was to provide a lecture on breast cancer diagnosis and the possible stages that might be found, which was seen "as a way to brace [themselves]" (Participant 8) for all diagnostic possibilities. The women recommended that they be asked if they would prefer to receive such a lecture while waiting for further testing or waiting for their results. Some of the women viewed the option of knowing in advance the possible implications of being diagnosed with cancer as a means by which to lower their uncertainties and distress.

In addition to their information needs, most women described a need for supportive care. The women described a need for preparatory emotional support or counselling to help acquire and build adequate coping skills prior to attending the RDC. The women suggested having a one-time telephone call from a nurse before and after attending an RDC to assess their support needs and coping skills. They also suggested having educational sessions on the upcoming procedures to reduce the uncertainties about the event on testing day. The women also recommend that prior to attending their full day of testing and waiting, someone from the clinic

should recommend they come with a support person. After all, as one woman shared, “we are here all day getting tests done, waiting for the results at the end of the day with the possibility of being told we have cancer. It is a lot to take in alone.”

5 | DISCUSSION

The study findings bring further insights to several current gaps in the empirical literature examining women’s emotional experiences of having a suspicious breast lump requiring further investigation through an RDC. As observed in this study, the women experienced high anxiety and uncertainty levels during the pre-diagnostic phase. These results accord with Mishel’s uncertainty theory (33) which explains that when faced with unfamiliarity, uncertainty arises. Thus, uncertainty theory would suggest that if women viewed the need for additional testing at the RDC as a threat rather than an opportunity, their appraisal of the situation would increase uncertainty and anxiety. In this study, the women faced the unfamiliarity of not knowing what to expect from their additional testing and faced a potential threat that could disrupt their daily lives. As explained by the uncertainty theory, being in an ill-defined situation further pulls individuals toward identifying the situation as a threat. When the event is viewed as a threat, there is a greater likelihood for an individual to experience higher anxiety levels. (11,34) All of these components likely played a role in the women’s emotional experience of undergoing further testing for a suspicious breast abnormality.

One avenue of research to explore further would be methods to guide and support those going through an RDC for suspicious breast abnormalities to assess their situation as an opportunity rather than a threat. Several examples of how the RDC experience could be framed as an opportunity include: 1) an opportunity for quick screening; 2) an opportunity to spend fewer days living with the uncertainty if their breast abnormality is cancerous or not, and in some instances; 3) an opportunity to benefit from a quicker turnaround to begin treatment.

By offering women optional perspectives to appraise their life-threatening situation positively, uncertainties experienced during the waiting period and upon the receipt of their diagnosis may be viewed instead as opportunities for action planning.

The mixed-methods approach in this study also led to further time-specific insights into the women’s emotional experiences. (35) The qualitative data helped explain the women’s reactions to the waiting period and the period following diagnosis. Within this group of women, a small group used positive reframing to keep their anxiety low while waiting to obtain further testing, while for others, the lack of understanding as to why the additional test was needed and how it would take place made them feel even more anxious and experience high levels of uncertainty. Harnessing quantitative evidence on the women’s emotional responses while also capturing the detailed nuances of their time-specific reactions via qualitative data helped achieve a more robust understanding of women’s experiences of rapid testing for breast abnormalities than with quantitative data alone.

The qualitative and quantitative results speak to a need for support to cope with the uncertain and highly anxious experience that comes from attending an RDC. Nurses are in an ideal position to provide and adopt this support to the psychological and educational needs of the individuals. One possibility to explore that may help decrease the short-term anxiety in individuals undergoing further cancer tests is for nurses to teach coping and relaxation skills such as positive reframing. Nurses could also facilitate communication with other professionals on the healthcare team and provide procedural support. (2,6,18,36,37) The integration of a nurse navigator in an RDC to reduce anxiety and increase satisfaction with care and services has been discussed positively in the literature. (38–40) Adapted education for RDCs with the support of a nurse navigator was reported as an important component to reducing distress and helping women prepare for decision-making around treatment options. (41)

5.1 | Implications for Practice

While waiting for further testing, most of the participants in this study experienced considerable heightened anxiety, and all scored above population norms for uncertainty. The participants expressed that these heightened levels impacted their daily personal, familial, and professional living activities. These results of high levels of anxiety are in accord with other studies investigating the emotional responses of women who received an abnormal screening mammography result and waiting for further testing and diagnosis. (42) For example, Pineault (42) noted that out of the 631 women who took part in their study, 51% were moderately or very anxious at every stage of the pre-diagnostic phase. Our results show that 69% of the women reported very high anxiety levels during the pre-diagnostic phase. Individuals attending RDCs are likely to experience clinical anxiety levels, and uncertainty warrants the consideration to screen for distress at the initial visit and each follow-up. While further studies are needed to assess equipoise between healthcare costs and enhanced patient experience with the implementation of distress screening, identifying individuals who may need further psycho-educational support can improve the quality of care and patient outcomes. (43) The findings further underline the need for emotional support during the post-testing period, especially for those found to have cancer. This study also suggests that at three weeks post-testing targeted psychosocial support may be helpful for those with a benign diagnosis.

The participants provided suggestions and several avenues on how to support those attending a rapid diagnostic clinic. One suggestion that aligns with the idea of a nurse navigator is to have this professional guide the day's process prior to and after attending the RDC, including guidance on the possible tests to be done. Several delivery options may need to be explored, such as group teaching, telehealth, or individual telephone support. (39) Considering the short time frame between the discovery of a suspicious breast lump to the day of testing at the RDC, the latter two support options might be most feasible. Patient preferences and individual needs

along with personnel availability may, in the end, dictate which approach would be most appropriate.

5.2 | Recommendations for Research

This study was explorative and a proof concept. There remain outstanding gaps in our understanding of the emotional impact and implications of attending an RDC after discovering a suspicious breast abnormality. Notably, future studies are needed to investigate how psycho-educational nursing-led interventions can reduce anxiety and uncertainty during and after the diagnostic process and which effective interventions provide patients with the greatest satisfaction. Such studies could guide the development of new models of care for RDCs across Canada. In addition, further studies could help identify risk factor profiles for high anxiety and uncertainty in women attending an RDC, such as individuals with a strong family history of the disease.

5.3 | Study Limitations

Like many other studies investigating the clinical and psychological impact of RDCs (16), this study's findings are constrained by its small sample size and with more than half of the sample having a family history with the disease. Given that this profile may not represent the general population, further research is needed to assess the associations between family history of cancer and anxiety levels and the need for genetic counselling and psychological support among those attending RDCs. This study was carried out in one cancer center offering rapid diagnostic testing for women with a suspicious breast lump, and the above profile may not represent other RDCs. However, the strengths of the mixed-methods findings contribute significantly to our empirical understanding of the women's emotional experience attending an RDC, and the potential role nurses can play within this clinical context to address the full range of emotional and information needs across all diagnosis types.

6 | CONCLUSION

RDCs offer women with a suspicious breast lump the opportunity for quicker diagnostic testing. The period, even if short, between the discovery of a suspicious breast abnormality and further testing at an RDC is marked by high, intense, emotional turmoil. Anxiety and uncertainty levels remain high for the group with malignant tumours post-diagnosis and three weeks post-diagnosis. Considering the women's descriptions and the anxiety and uncertainty levels observed by the women who took part in this study, further investigation towards the possible provision of psychosocial support as part of the nurse working in RDC care is warranted.

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8 | CONFLICT OF INTEREST

The authors have no conflicts of interest to declare. All authors have made substantial contributions and had full access to the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. There is no financial interest to report.

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Strategies to Help Mothers of Neonatal Intensive Care Unit Infants Cope with Milk Expression

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COMMENTARY ON

Bujold, M., Feeley, N., Axelin, A., Cinquino, C., Dowling, D., Thibeau, S. Expressing Human Milk in the NICU. 2018. *Advances in Neonatal Care*; 18(1): 38-48.

SUMMARY

Efforts to adopt the national breastfeeding recommendations in Canada, which recommend exclusive human milk consumption, (1) present many challenges for mothers who have to express their milk for weeks or months while their infant is in the neonatal intensive care unit (NICU). Bujold et al. conducted a descriptive, qualitative cross-sectional study to explore these challenges and offer solutions to foster maternal feelings of infant closeness and motivation to express milk. (2) An infographic depicting positive coping strategies was created to optimize the practice of neonatal nurses in the NICU and help them address these challenges.

KEYWORDS

NICU, Milk expression, Coping, Breastfeeding

1 | IMPLICATIONS FOR PRACTICE AND RESEARCH

Fluctuations between feelings of closeness to and separation from an infant play an important role in milk expression for mothers whose infants are in the NICU. Positive coping strategies must be promoted by neonatal nurses, such as encouraging support-seeking or expressing milk during or right after skin-to-skin contact. (2)

2 | CONTEXT

Although human milk consumption has many health benefits for newborns, mothers of preterm infants can face many challenges with expressing milk for an extended period of time while their infant is in the NICU. (2) Bujold et al. sought to document these challenges, explore how mothers cope with them, and determine if expressing milk can lead to feelings of closeness or sep-

aration from their infant.

3 | METHODS

Bujold et al. conducted a descriptive, qualitative cross-sectional study to explore the personal experiences of mothers expressing milk in a NICU setting. Fifteen mothers were provided with a smartphone equipped with an application allowing them to document their experience. Each time they expressed milk over a 48-hour period, mothers were asked to indicate whether the episode led to a feeling of closeness or separation, as well as audio-record their thoughts and feelings. Data was analyzed using a thematic content analysis. (2)

4 | FINDINGS

Feelings of closeness and separation experienced by mothers during milk expression were influenced by challenges, coping strategies, location and environment. (2) Mothers who coped with the difficulties associated with milk expression by using various strategies reported a more positive bond with their infant compared to mothers who were overwhelmed. (2) Mothers more frequently reported feeling separated from their infant when they expressed milk at home compared to when at their infant's bedside or in a NICU milk expression room. (2)

5 | COMMENTARY

Consumption of human milk helps reduce neonatal mortality. (3) It may also help address mothers' mental health as postpartum depression may be linked to the absence or early cessation of breastfeeding. (4) Bujold et al. suggest that promoting positive strategies to cope with milk expression may help foster maternal feelings of closeness with their infant and therefore increase breastfeeding rate in the NICU. (2) For example, skin-to-skin contact reinforces feelings of closeness, physiologically reduces stress and increases milk supply. (5)

Similarly, music therapy was shown to reduce cortisol levels and significantly increase the amount of milk expressed. (6) Other effective distractions, such as reading a book, should be considered on an individual basis to positively change the mother's mindset while pumping. The addition of peer counsellors in lactation programs was shown to increase the rate of exclusive human milk consumption in the NICU and even at discharge. (7) Offering positive reinforcement, encouraging mothers to seek support from significant others, and establishing a peer-to-peer mentoring program would help address the challenges faced by mothers expressing milk while their infant is in the NICU. Several other strategies (**Figure 1**) can be encouraged by neonatal nurses in an effort to reinforce feelings of closeness, help mothers cope with milk expression and increase human milk consumption in the NICU. The successful implementation of these initiatives by nurses would benefit from a standardized guideline, as proposed in **Figure 1**.

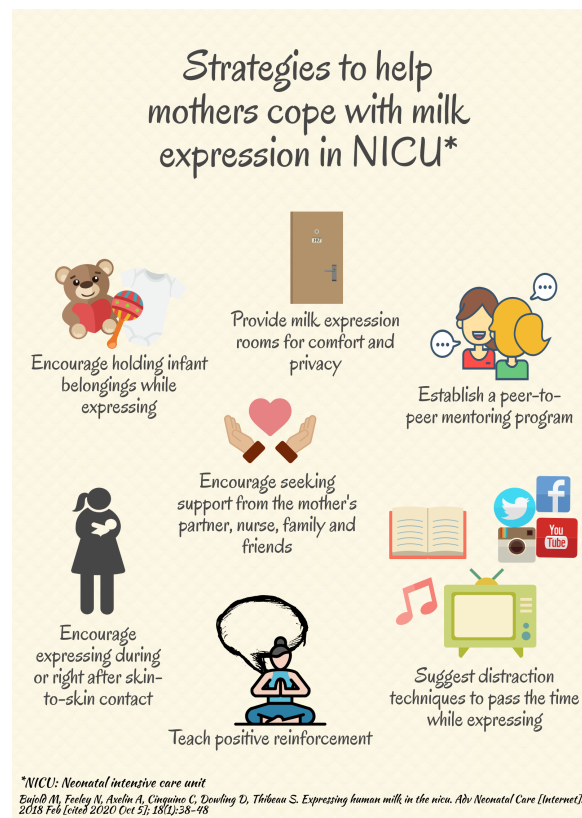


FIGURE 1 Strategies to help mothers cope with milk expression in NICU

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Implementing Telehealth Services with the B.E.L.T.™ Tool: A Commentary

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COMMENTARY ON

B.E.L.T.™: Arnaert A, Debe Z. BELT™: Framework for Nurse Champions to Successful Implement Sustainable Telehealth Services. *Iris Journal of Nursing and Care* [Internet]. 2019 [cited 2020 nov 25];1(5). Available from: <https://pdfs.semanticscholar.org/3659/c19d1c2d19cf35fee334b4fd261790edc934.pdf>

SUMMARY

As technology takes on an increasingly important role in our everyday lives, the concept of telehealth is taking form in healthcare. Indeed, evidence shows that it can facilitate and improve access to quality care. This commentary offers a synopsis on the development of the B.E.L.T. mnemonic, to be used by nurse champions to structure the implementation of telehealth services. Later research indicates that additional testing is needed to adapt and further validate the tool. The COVID-19 worldwide pandemic has provided the ideal opportunity to investigate this tool given the widespread and rapid implementation of telehealth in different settings worldwide.



KEYWORDS

B.E.L.T.™, Telehealth implementation, Nursing framework

1 | IMPLICATIONS FOR PRACTICE AND RESEARCH

- Use the B.E.L.T. framework to facilitate the use and uptake of telehealth services.
- The worldwide COVID-19 pandemic indicates the need for a quick systematic approach to implementation of telehealth services across healthcare disci-

plines.

2 | CONTEXT

Technology is widely used in health care and allows for improved access to care with minimal cost and need for travel. Putting in place a telehealth system and adapt-

ing it to a specific clinical context may be complex and require a systematic approach. Therefore, the use of a framework would be beneficial for nurse champions undertaking such implementation. (1)

3 | METHODS

Drawing from their previous telehealth experiences, Arnaert and Debe (2019) share their narrative on the development of the B.E.L.T. framework to complement existing guidelines and recommendations and showcase its application for the provision of telehealth services. Arnaert and Debe (2019) discuss the successes and challenges encountered when usually launching telehealth services and from these delineate the core elements necessary for a health care organization to achieve and sustain telehealth services.

4 | FINDINGS

The B.E.L.T. mnemonic integrates these core elements into a “checklist” for nurses establishing telehealth services. The mnemonic is as follows:

B: Bandwidth/Broadband: Assess appropriate internet connection and speed needed to ensure uninterrupted transmission of information.

E: Education/Environment: Develop protocols and tailor the education according to the socio-cultural environment of the patients and professionals.

L: Leadership: Assign a nurse champion to be the leader, coordinator and advocate.

T: Technology: Assess the variety of technology and available infrastructures for implementing telehealth.

This mnemonic has been summarized as an infographic by the author of this commentary to encourage use (Figure 1).

5 | COMMENTARY

The B.E.L.T. framework complements the existing guidelines for telehealth implementation and serves as an

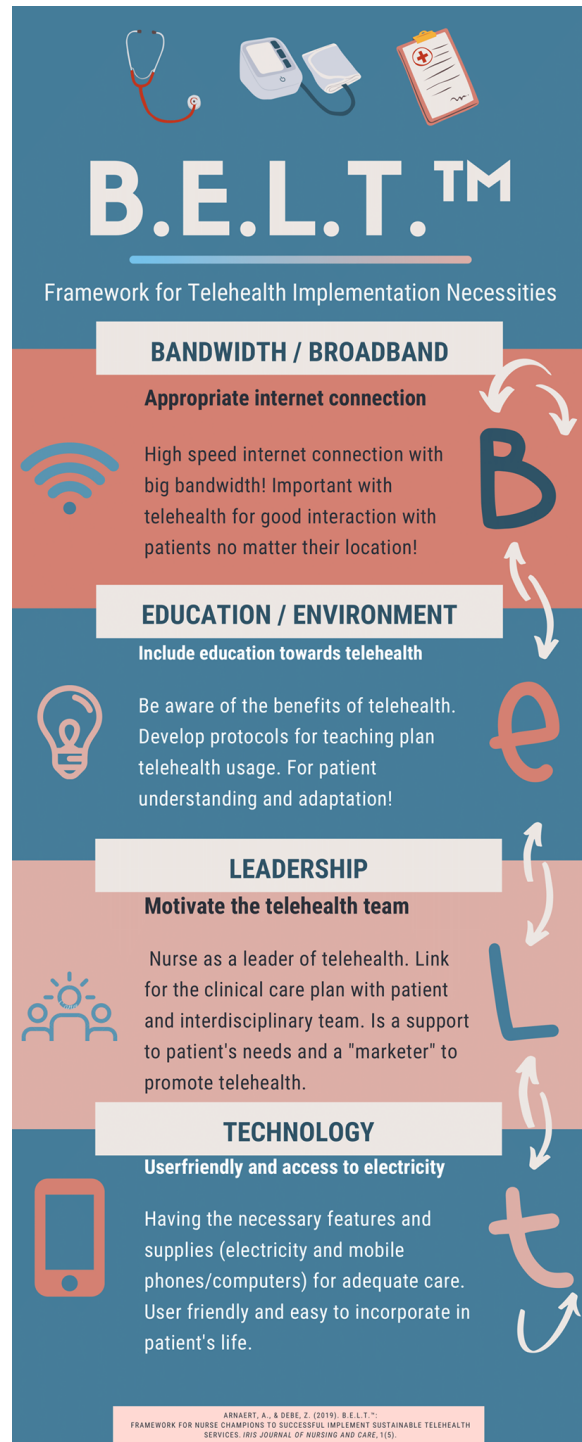


FIGURE 1 Infographic of the B.E.L.T.™ Framework

easy-to-use checklist for nurses to facilitate the use and uptake of telehealth services in health care organizations. (1) Arnaert et al (2019) used the BELT frame-

work to implement a telehealth pilot project for screening and monitoring pregnant women in Rural Burkina Faso, Africa. While the mnemonic served as the initial step to guide the implementation of this program, Arnaert et al (2019) identified other factors to consider such as conducting an assessment of the region's rural context and demographics prior to implementation. (2) Within the current context of the COVID-19 pandemic, person-to-person contact should be avoided when possible. The utilization of telehealth services has therefore increased since March 2020 and has become imperative to consultations with non-urgent clients as well as meetings with caregivers unable to accompany hospitalized/institutionalized patients. (3, 4) Finally, although the B.E.L.T. framework can be an essential tool for developing and implementing telehealth services, further research is necessary to validate this framework in other clinical settings.

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Osteogenesis Imperfecta (OI) Transfer Summary Tool May be Used to Facilitate the Transfer of OI Patients from the Pediatric to Adult Healthcare Systems

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COMMENTARY ON

Carrier J, Siedlikowski M, Chougui K, Plourde S-A, Mercier C, Thevasagayam G, et al. A best practice initiative to optimize transfer of young adults with osteogenesis imperfecta from child to adult healthcare services. *Clin Nurse Spec.* 2018 Nov/Dec;32(6):323-35. Available from: <https://pubmed.ncbi.nlm.nih.gov/30299335/> doi: 10.1097/NUR.0000000000000407

SUMMARY

What happens once youth and young adults with osteogenesis imperfecta (OI) are transferred from the pediatric to the adult healthcare system? To facilitate this process, an interprofessional task force was convened to conduct a knowledge synthesis study, which entailed reviewing the literature, developing guidelines and creating a transfer tool. This study and later research indicate the need for the creation of a transitional care program tailored to the special needs of OI patients and their families.

KEYWORDS

Osteogenesis Imperfecta, Transfer tool, Transition, Health care system

1 | IMPLICATIONS FOR PRACTICE AND RESEARCH

- Integrate the use of the OI Transfer Tool to facilitate the transfer of youth and young adults from the pe-

diatric to the adult care health care systems.

- Create a transitional care program to assist patients and families in the transition process and provide adult healthcare practitioners with the key information and tools to care for youth and young adults.

2 | CONTEXT

Similar to other young adults with a chronic illness, (2) the transition to the adult healthcare system is complex for youth and young adults with OI who have expressed concerns with their transition. (3) The multi-faceted nature of their rare disease, paucity of evidence of research, and absence of tools to facilitate the transfer further compound the issue. (4) This places these patients at heighten risk for complications. (4) Hence, an inter-professional Task Force with international expertise in OI including patient representation was convened to address the notable gap in the transfer of youth and young adults with OI to the adult health care system.

3 | METHODS

Carrier et al. led an interprofessional expert task force at a university-affiliated hospital with international expertise in OI to: 1) review the literature, 2) develop guidelines, and 3) create a tool to facilitate the transfer of youth and young adults with OI from the pediatric to the adult health care systems. Six electronic databases and varying grey sources—literature not published through traditional means—from the US and Canada were searched. Further contact was made with transition program coordinators in Canada or the United States to retrieve published and unpublished transfer tools. All data were extracted, descriptively summarized, and appraised by the Task Force who had no conflicts of interest.

4 | FINDINGS

Carrier et al (2018) reported a paucity of research in this area. After a comprehensive literature review, only 7 studies were identified. Eight transfer tools were retrieved and descriptively summarized. Together, with the paucity of evidence, the Task Force relied on their interprofessional expertise in OI to create the “OI Transfer Summary” tool. This tool comprised 11 sections summarizing the care given in the pediatric health care system,

and the follow-up requirements. Therefore, a whole overview of the patient is presented to the new caregivers and offers a starting point for the provision of individualized primary care in the adult health care system.

5 | COMMENTARY

Transferring from pediatric to adult systems may be a challenging event in lives of youth and young adults with OI. (3) To date, clinicians and researchers support the creation of a transitional care program to guide youth and young adults with OI and their families to the adult health system. (3) This program would furthermore support the clinicians in the adult sector to receive the transfer and attend to their health care needs. (3) The OI Transfer Summary tool, along other existing OI passports, (5, 6) may be incorporated into such a program to offer personalized care and optimize communication between transfers. (7, 8) Derived from existing transitional care programs, (9, 10) this program may include emphasis on familiarizing patients with the adult system, such as preparing for adult care provider appointments, visiting clinics and precocious preparation, and including peer mentorship. (3, 8, 9, 11) The OI Transfer Tool would serve as a gateway to facilitate the discussion of these items as the patient completes it in collaboration with the interprofessional team. Moreover, the tool may help prompt educating patients on their responsibilities and assessing for their transfers readiness, (12) which collectively may ease the transition process. However, further research is needed as this tool has not yet been evaluated in practice, and the outcomes of using such a tool are unknown. Nevertheless, ideal practices suggest that bridging the transition process and optimizing the knowledge gap among adult healthcare professionals, a transfer coordinator, and integration of technology could be used to educate and facilitate their access to resources in person and online. (13)

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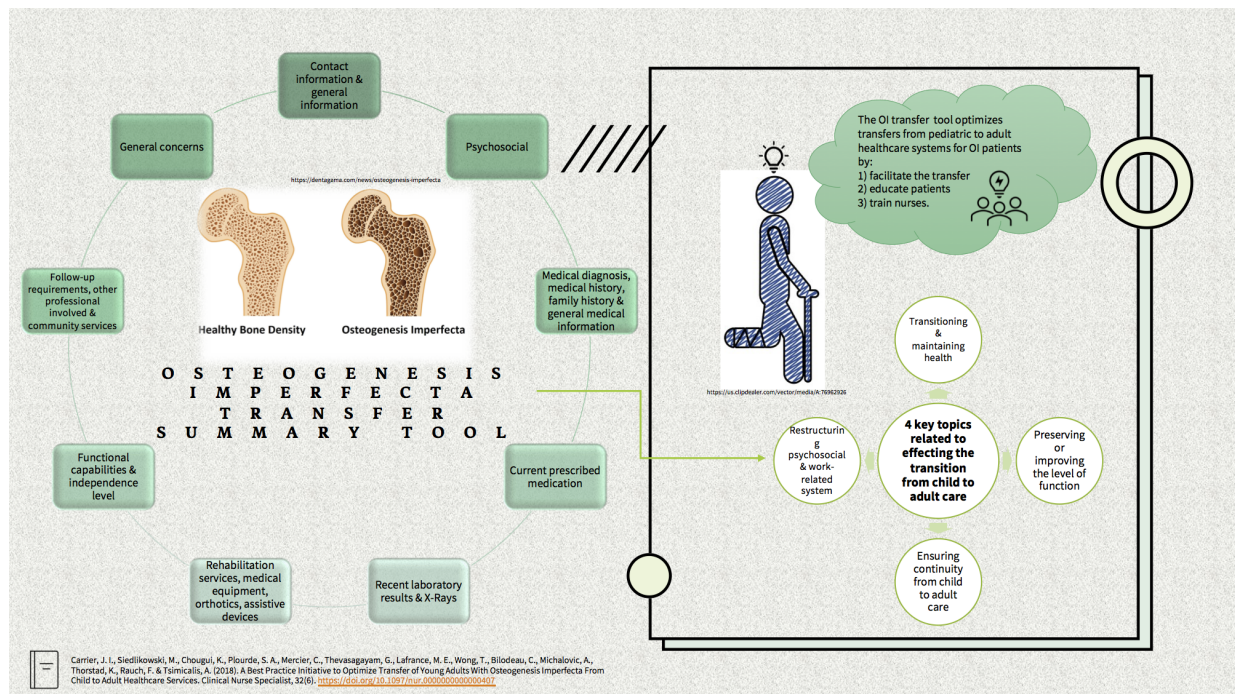


FIGURE 1 An Infographic Summary of the Key Components of the OI Transfer Summary Tool and Objectives Guiding its Creation (1).

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BRIEF REPORT

McGill Journal of Medicine

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 stairlifts. 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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