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Digital Tools to Support Family-Based Weight Management for Children: Mixed Methods Pilot and Feasibility Study

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Abstract

Background: Family-based behavioral therapy is an efficacious approach to deliver weight management counseling to children and their parents. However, most families do not have access to in-person, evidence-based treatment. We previously developed and tested DRIVE (Developing Relationships that Include Values of Eating and Exercise), a home-based parent training program to maintain body weight among children at risk for obesity, with the intent to eventually disseminate it nationally alongside SafeCare, a parent support program that focuses on parent-child interactions. Currently the DRIVE program has only been tested independently of SafeCare. This study created the “mHealth DRIVE” program by further adapting DRIVE to incorporate digital and mobile health tools, including remotely delivered sessions, a wireless scale that enabled a child-tailored weight graph, and a pedometer. Telehealth delivery via mHealth platforms and other digital tools can improve program cost-effectiveness, deliver long-term care, and directly support both families and care providers.

Objective: The objective of this study was to examine preliminary acceptability and effectiveness of the mHealth DRIVE program among children and parents who received it and among SafeCare providers who potentially could deliver it.

Methods: Study 1 was a 13-week pilot study of a remotely delivered mHealth family-based weight management program. Satisfaction surveys were administered, and height and weight were measured pre- and post-study. Study 2 was a feasibility/acceptability survey administered to SafeCare providers.

Results: Parental and child satisfaction (mean of 4.9/6.0 and 3.8/5.0, respectively) were high, and children’s (N=10) BMI z-scores significantly decreased (mean –0.14, SD 0.17; P=.025). Over 90% of SafeCare providers (N=74) indicated that SafeCare families would benefit from learning how to eat healthily and be more active, and 80% of providers reported that they and the families would benefit from digital tools to support child weight management.

Conclusions: Pediatric mHealth weight management interventions show promise for effectiveness and acceptability by families and providers.


KEYWORDS
parent training; weight loss; telehealth; obesity; SafeCare
Introduction

Obesity affects nearly one in five children and adolescents in the US [1]. The US Preventive Services Task Force [2] and the American Medical Association [3] recommend comprehensive, intensive, family-based weight management programs to treat childhood obesity. Family-based behavioral therapy is efficacious [4], although most children do not have access to evidence-based treatment due to limited availability of programs and trained providers, barriers for travelling to in-person sessions including transportation and time constraints, and cost of participation due to limited or no insurance coverage [5,6].

To overcome barriers to access, evidence-based models that include parent training (eg, SafeCare, Parents as Teachers) can be delivered in the family’s home [7]. SafeCare is a parent support program delivered by trained providers that focuses on parent-child interactions to mitigate the risk of abuse or neglect. SafeCare is predominantly delivered in the home, but sessions can also be delivered via technology, over video chat and telephone [8]. SafeCare has been disseminated in more than 25 US states and internationally. Currently, there are approximately 100 SafeCare accredited agencies where providers serve more than 6000 families per year. The underlying principles of SafeCare on improving parent-child interaction, coupled with its broad reach to at-risk and underserved families, make SafeCare an ideal platform for delivery of weight management services.

We developed a parent support focused program to treat childhood obesity that can be delivered in the home called DRIVE (Developing Relationships that Include Values of Eating and Exercise) [9] with the intent to eventually disseminate the program across the SafeCare network. DRIVE incorporates SafeCare principles to promote healthy eating, physical activity, and healthy weight in children by fostering positive parent-child interactions. Previously, we tested the efficacy of DRIVE in a 19-week randomized controlled pilot trial in 16 parent/child dyads (children ages 2-6 years with BMIs ≥ 75th percentile) and found that the change in children’s BMI z-scores (BMIz) (Mean –0.1, SE 0.1) was significantly different (P<.01) compared to a health education control group (mean 0.5, SE 0.1) [9].

Although DRIVE was initially developed for in-person delivery, telehealth delivery via mHealth platforms can improve cost-effectiveness, deliver long-term care, and directly support both families and care providers [10]. Identifying alternate avenues for families to access care is increasingly important [10], including for children with obesity during the COVID-19 pandemic when families are unwilling or unable to present in-person for treatment [11]. To this end, the objectives of the studies reported herein were to examine 1) the acceptability of a remotely delivered weight management program (mHealth DRIVE) as determined by the parents and children who used the program, 2) the preliminary effectiveness of this virtual program to reduce child body mass, and 3) the perceived need and willingness to deliver mHealth DRIVE by SafeCare providers.

Methods

Study 1

Participants

Parents were recruited from their children’s after-school wellness program. Parents were invited to attend an informational session that explained the purpose of mHealth DRIVE. Eligibility criteria for children included ages 5 to 14 years; be physically capable of exercise; and be free of diseases that affect metabolism, body weight, and food intake, including type 1 or type 2 diabetes, HIV/AIDS, and cancer. Children were excluded if they had significant cardiovascular disease or disorders or other significant medical problems that would prevent them from engaging in regular physical activity. Inclusion criteria for parents included having a smart phone and being willing to use the smartphone for the intervention. Eleven child/parent dyads enrolled, but 1 dyad was excluded from all analyses because of the child’s low BMI percentile (4th percentile). Parents provided written informed consent, and children provided assent. Study procedures were approved by the Pennington Biomedical Research Center institutional review board.

Intervention Sessions, Treatment Goals, and Tracking of Weight and Behaviors

Child/parent dyads attended 8 counseling sessions (approximately 30 minutes each) primarily over their internet-connected device (eg, smartphone, tablet, laptop, or desktop computer). Most interactions were via video calls or phone, but email and text communication also occurred. The DRIVE curriculum was shortened to 13-weeks to align with the school semester. A Pennington Biomedical counselor delivered sessions and provided individualized advice and problem-solving strategies for the parent and the child. Each session included an interactive component for the parent and child related to healthy eating and active play, and interactive parenting training. Sessions were based on treatment methods that promote child weight loss that have been sustained for 10 years [12,13]. Although the sessions were remotely delivered, counselors were able to deploy motivational interviewing techniques to address decreases in motivation, which are inevitable in longer-term interventions [14].

The guiding principles of the sessions were 1) weight and activity monitoring; 2) building commitment and overcoming barriers to healthy behavior changes, with a goal of teaching the parent to model appropriate diet and physical activity behaviors for their child; 3) review of progress and problem-solving to address poor adherence to behavioral goals; and 4) food monitoring and goal setting for nutrient intake. Sessions focused on how to motivate the child and manage noncompliance; techniques included praise and reward, positive reinforcement, selective ignoring, contracting, preplanning for meals and physical activity, shaping behaviors, modeling, changes to the home environment, and facilitating social support for behavior change [15,16]. The dietary approach employed food monitoring and goal setting for nutrient intake, and the Traffic Light Diet [12] was included to facilitate remote
modification of dietary changes. The Traffic Light Diet teaches parents and children to categorize foods based on green (low calorie foods to be eaten frequently), yellow (moderate-calorie foods to be eaten occasionally), and red (high-calorie foods to be eaten rarely), with the goal to gradually reduce the number of red foods eaten each week. The physical activity approach introduced free or inexpensive activity options that the children enjoyed and addressed barriers to physical activity.

Children’s energy requirements were estimated using a physical activity level of 1.4 and the Harris-Benedict equation, which has good accuracy in youth with obesity [17]. The energy intake goal was 250 kcal/d less than estimated energy requirements, which should promote modest weight loss and weight gain attenuation over time. The activity goal of children was to gradually increase physical activity to a goal of approximately 6,000 steps/day above their personal baseline values, which is appropriate as we expected low baseline physical activity [18]. This activity goal was the equivalent of an additional 30 min/day of moderate-to-vigorous physical activity as a gradual increase towards the physical activity guidelines of 60 min/day of moderate-to-vigorous physical activity [18].

The intervention content and parent-training approach were based on DRIVE, and the mHealth aspects of the intervention were based on a successful weight management intervention for adults called SmartLoss [19,20]. Specifically, children’s daily physical activity (steps/day) was tracked with a hip-worn Omron HJ-324U pedometer (Omron Healthcare, Inc, Kyoto, Japan), and the parent was asked to document their child’s steps daily. The counselor plotted the child’s daily step data in relation to their individual goals to help promote adherence to activity goals. The children also received a BodyTrace scale that automatically sent their weights to a website accessible by the counselors. Children were asked to weigh themselves at least weekly, unless contraindicated due to anxiety or other mental health barriers, similar to SmartLoss [19,20]. Weighing at the same time of day and in the same state was encouraged, preferably after getting out of bed in the morning and after voiding. Children’s body weight and a weight graph were used to guide intervention delivery to facilitate healthy weight management and avoid unsafe changes in body weight. Specifically, and as detailed in the upper panel of Figure 1, a 6-pound “zone” of acceptable weights or “adherence” was created, and children’s individual weights were plotted against this zone. Hence, the zone promotes weight maintenance, but it allows for weight loss of less than 3 pounds if the child’s BMI is greater than or equal to the 85th percentile. Further, this approach includes objective safety criteria that are triggered if rapid or excessive weight loss occurs (see Figure 1, lower panel).

The program encourages healthy eating, activity, and weight tracking over time. The counselor and parent utilized the weight graph to modulate intervention intensity and as an objective indicator of the need to change the child’s energy intake level. Specifically, the counselor and parent 1) increase energy intake if weight loss is excessive, defined as more than 0.2 BMIz reduction within 1 month, which aligns with American Medical Association recommendations for maximum 2 lb/week weight loss in children [3]; 2) maintain energy intake if weight maintenance is observed, until the child’s BMIz reaches 0.25 (approximately equivalent to the 60th BMI percentile); and 3) reduce energy intake if the child is gaining weight at a rate that increases the child’s BMIz, unless he/she has reached 0.25 BMIz or approximately the 60th percentile, at which time the child increases body weight over time to maintain 0.25 BMIz or approximately the 60th percentile. The threshold of 0.25 BMIz to begin weight maintenance aligns with the goal of reducing BMIz without promoting energy restriction that could negatively impact growth and development. In a longer-term intervention, the zone would be adjusted every 6 months according to increases in the child’s height (see Figure 1), but this pilot study did not adjust the zone due to the study being only 13 weeks in duration. The counselor electronically provided the parent with the child’s weight graph during each session (see the upper panel of Figure 1).

Parents who needed help modifying their child’s diet had the option of sending their counselor images of how they prepare foods and what foods they provide to their child and family. These were not outcome data but provided the counselor with near real-time data on changes the parents could make to improve their child’s diet and health. These images can be captured with any camera-enabled device, and smartphone apps are available to streamline this process (eg, SmartIntake).

Figure 1. Weight graph zone of child’s adherence. BMIz: BMI z-score.
Measures and Data Analysis

Parents and children completed an acceptability survey at the end of the intervention that included Likert scales on intervention satisfaction (see Table 1). Children’s height and weight (shoes removed, no outer clothing) were collected in duplicate at baseline and end of study by trained assessors, and both assessments occurred in the afternoon. Height was measured with a stadiometer, with the child standing feet flat, with heels, buttocks, upper back, and back of head contacting the stadiometer, and the child’s head facing straight ahead. Weight was measured with a digital scale with the child standing in the middle of the scale with arms hanging loosely at their side. Height and weight were recorded to the nearest 0.1 unit (cm or kg, respectively); if the two measures differed by more than 0.5 units, a third measurement was taken and the closest two of three were used in analysis. Mean values and percentages were calculated for satisfaction surveys. Differences in BMI were examined using t-tests, with an alpha level of .05. Analyses were conducted using SPSS.

Study 2

A survey of SafeCare providers was conducted across the US to assess 1) the perceived need for diet, physical activity, and weight management services for SafeCare children, and 2) the willingness of SafeCare providers to offer such services.

Participants

Eighty-two SafeCare providers from 14 states provided consent and completed the survey. The sample was predominantly female (n=71), with 5 males, 1 other, and 5 unknown. The mean age of providers was 39.8 years (SD 12.9), with 17 unknown age data. Thirty-eight providers reported delivering care in urban cluster/suburban areas (2500-50,000 people), 22 in urban areas (≥50,000 people), and 18 in rural areas (<2,500 people), with 4 unknown.

Procedures

A recruitment email was sent to the potential participants using the list of contact information for US SafeCare providers. The email contained an anonymous link to the survey conducted through Qualtrics, a secure web-based survey platform that employs high-level security measures to ensure data are protected from malicious data breaches and requires a password in order to download the data. A reminder email was sent 1 week later, reminding participants of the opportunity to complete the survey. The survey was open for 2 weeks.

Measures and Data Analysis

The survey queried demographic data (age, gender, and level of urbanicity where services are delivered) and assessed if SafeCare providers perceive a need for or have experience with additional educational material for child nutrition/weight management. Data were cross-sectional and were analyzed descriptively (ie, percentages were reported for categorical variables; means or percentages were reported for Likert scale items). Reported percentages collapse the “Strongly Agree” and “Agree” responses.

Results

Study 1

Of the 10 children, 6 were girls (60%) and the mean age was 7.8 years (SD 2.3 years; range 6-14 years). Mean BMI percentile and BMIz were 86th (SD 0.17) and 1.4 (SD 0.7), respectively. Four children had obesity, 4 were overweight, and 2 were normal weight. There was a statistically significant reduction in children’s BMIz over the 13-week period (mean –0.14, SD 0.17; P=.025). There was also a significant BMIz reduction among the 8 children who were overweight or had obesity (mean –0.18, SD 0.15; P=.013). The 2 normal weight children did not lose weight. Parental satisfaction (4.9/6.0) and child satisfaction (3.8/5.0) were high (see Table 1).
Table 1. Parent (n=10) and child (n=10) satisfaction survey results.

<table>
<thead>
<tr>
<th>Survey items</th>
<th>Rating score</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Parent items 1 (responses ranged from 1=Strongly Disagree to 6=Strongly Agree)</strong></td>
<td></td>
</tr>
<tr>
<td>Seeing my child’s weight on a graph every week helped me make better food choices for him/her.</td>
<td>4.4 (1.2)</td>
</tr>
<tr>
<td>My child was willing to step on the bathroom scale once per week.</td>
<td>5.6 (0.8)</td>
</tr>
<tr>
<td>My child was willing to wear a pedometer every day.</td>
<td>4.0 (2.4)</td>
</tr>
<tr>
<td>Tracking my child’s steps each day helped him/her reach physical activity goals.</td>
<td>4.5 (1.7)</td>
</tr>
<tr>
<td>Tracking the foods my child ate helped him/her reach weight goals.</td>
<td>5.1 (1.2)</td>
</tr>
<tr>
<td>The healthy tips my child and I received helped me make healthy lifestyle changes for my child.</td>
<td>5.4 (0.9)</td>
</tr>
<tr>
<td>The information I received in my health tips helped me make healthy lifestyle changes for my family &amp; myself.</td>
<td>5.3 (0.8)</td>
</tr>
<tr>
<td>I enjoyed the individual time talking with my counselor.</td>
<td>5.8 (0.4)</td>
</tr>
<tr>
<td>The amount of time talking with my interventionist was enough.</td>
<td>5.7 (0.5)</td>
</tr>
<tr>
<td>I would have liked to spend more time talking with my interventionist.</td>
<td>1.8 (0.9)</td>
</tr>
<tr>
<td>I enjoyed meeting with my counselor remotely (by phone call or video chat on my smartphone).</td>
<td>5.8 (0.4)</td>
</tr>
<tr>
<td><strong>Parent items 2 (responses ranged from 1=Not helpful to 6=Very Helpful)</strong></td>
<td></td>
</tr>
<tr>
<td>Learning about the importance of self-monitoring how much we eat and our activity.</td>
<td>5.7 (0.5)</td>
</tr>
<tr>
<td>Learning about portion control.</td>
<td>5.7 (0.5)</td>
</tr>
<tr>
<td>Learning about choosing the right foods for you and your child.</td>
<td>5.4 (0.7)</td>
</tr>
<tr>
<td>Learning about how to build good social support.</td>
<td>5.4 (0.9)</td>
</tr>
<tr>
<td>Learning about fat, protein, and carbohydrates.</td>
<td>5.5 (0.5)</td>
</tr>
<tr>
<td>Learning about how to overcome barriers to being healthy.</td>
<td>5.4 (0.9)</td>
</tr>
<tr>
<td>Learning about how to make better choices when eating outside the home.</td>
<td>5.7 (0.5)</td>
</tr>
<tr>
<td>Learning how to make healthy choices on special occasions such as birthday parties and school functions.</td>
<td>5.3 (0.6)</td>
</tr>
<tr>
<td>Learning about healthy eating plans for the whole family, like the Stoplight approach to healthy eating.</td>
<td>5.7 (0.5)</td>
</tr>
<tr>
<td>Learning about how much physical activity is recommended for me and my child.</td>
<td>5.5 (0.5)</td>
</tr>
<tr>
<td>Taking a closer look at why we eat.</td>
<td>5.3 (0.6)</td>
</tr>
<tr>
<td>Learning about healthy beverage choices for me and my child.</td>
<td>5.2 (1.5)</td>
</tr>
<tr>
<td><strong>Child items (responses were 1=No; 2=I don’t think so; 3=Maybe; 4=I think so; 5=Yes)</strong></td>
<td></td>
</tr>
<tr>
<td>I liked wearing my pedometer.</td>
<td>3.1 (0.4)</td>
</tr>
<tr>
<td>I liked seeing how many steps I can get each day.</td>
<td>4.3 (0.3)</td>
</tr>
<tr>
<td>The pedometer was easy to use.</td>
<td>3.5 (0.5)</td>
</tr>
<tr>
<td>I tried to move more.</td>
<td>3.5 (0.5)</td>
</tr>
<tr>
<td>I liked talking to [interventionist] about eating healthy foods and being more active.</td>
<td>4.0 (0.4)</td>
</tr>
<tr>
<td>I tried to eat healthier foods.</td>
<td>4.1 (0.4)</td>
</tr>
<tr>
<td>I tried new healthy foods that I had not tried before.</td>
<td>3.8 (0.5)</td>
</tr>
<tr>
<td>I ate less candy.</td>
<td>3.3 (0.5)</td>
</tr>
</tbody>
</table>
Study 2
Nearly all respondents indicated that SafeCare families would benefit from learning how to eat more healthily and be more active (71/74, 96% and 68/74, 92%, respectively), and many (57/72, 79%) perceived that families would benefit from a program for child weight management. Most providers indicated that they were interested in learning how to deliver nutrition and physical activity information to their families (70/74, 95% and 60/74, 81%, respectively). About 80% (59/74) of providers reported that they and their SafeCare families would benefit from digital tools to support child weight management (see Table 2).

Table 2. Mean feasibility ratings reported by SafeCare providers (N=74), followed by the number (n) of providers who endorsed each rating from Strongly Disagree (1) to Strongly Agree (4).

<table>
<thead>
<tr>
<th>Survey items</th>
<th>Mean (SD)</th>
<th>Strongly Disagree, n</th>
<th>Disagree, n</th>
<th>Agree, n</th>
<th>Strongly Agree, n</th>
</tr>
</thead>
<tbody>
<tr>
<td>The parents I work with have regular access to healthy foods.</td>
<td>2.5 (0.7)</td>
<td>7</td>
<td>25</td>
<td>39</td>
<td>3</td>
</tr>
<tr>
<td>The parents I work with and their families would benefit from learning more about how to eat healthy.</td>
<td>3.3 (0.6)</td>
<td>1</td>
<td>2</td>
<td>44</td>
<td>27</td>
</tr>
<tr>
<td>I would be interested in learning how to deliver nutrition information to the parents I work with.</td>
<td>3.4 (0.6)</td>
<td>0</td>
<td>4</td>
<td>38</td>
<td>32</td>
</tr>
<tr>
<td>Most of the parents I work with or their families would benefit from weight loss or better weight management.</td>
<td>2.9 (0.8)</td>
<td>1</td>
<td>25</td>
<td>28</td>
<td>20</td>
</tr>
<tr>
<td>I would be interested in learning how to deliver weight management information to the parents I work with.</td>
<td>2.9 (0.9)</td>
<td>5</td>
<td>20</td>
<td>25</td>
<td>24</td>
</tr>
<tr>
<td>The parents I work with and their families would benefit from learning more about healthy levels of physical activity and exercise.</td>
<td>3.3 (0.6)</td>
<td>0</td>
<td>6</td>
<td>43</td>
<td>25</td>
</tr>
<tr>
<td>I would be interested in learning how to deliver information on physical activity to the parents I work with.</td>
<td>3.1 (0.7)</td>
<td>0</td>
<td>14</td>
<td>38</td>
<td>22</td>
</tr>
<tr>
<td>The parents I work with would benefit from a home visiting program designed to improve the body weight and health of young children in the home.</td>
<td>3.0 (0.7)</td>
<td>1</td>
<td>14</td>
<td>40</td>
<td>17</td>
</tr>
<tr>
<td>The parents I work with would benefit from mobile health tools (smartphones, online dashboards) designed to improve their diet, activity levels, body weight, and health.</td>
<td>3.1 (0.7)</td>
<td>0</td>
<td>14</td>
<td>42</td>
<td>18</td>
</tr>
<tr>
<td>I would be interested in receiving support via mobile health tools (smartphones, online dashboards) to help me deliver health and weight management information to the parents I work with and their families.</td>
<td>3.1 (0.7)</td>
<td>0</td>
<td>15</td>
<td>34</td>
<td>25</td>
</tr>
</tbody>
</table>

aN=72 due to missing responses.
Discussion

Principal Findings

In this one-arm small pilot study, an mHealth weight management program significantly reduced children’s BMIz, and both parents and children had high levels of satisfaction. These data complement and build upon the prior DRIVE in-person home-based weight management program by integrating digital tools including telehealth counseling sessions, a wireless scale that enabled a child-tailored weight graph, and a pedometer to track child physical activity. Further, the survey of SafeCare providers indicated that providers perceive a need for this type of family-based weight management program and expect that their families will find remotely delivered content and digital tools to be acceptable.

Collectively, these preliminary data suggest that a weight management program delivered to parent/child dyads may be successful when implemented alongside a parenting program, such as SafeCare, via an mHealth platform. These data contribute to the burgeoning evidence that telehealth may be useful as adjunctive to in-person pediatric weight management. A nonrandomized comparative effectiveness study of 100 adolescents participating in a 2-year weight management program compared in-person plus telehealth versus in-person only and observed similar BMI outcomes, attendance rates, and acceptability among families and healthcare providers across the two groups [21]. Digital tools may not only remove barriers to transportation and scheduling for in-person care delivery but also expand reach of interventions to areas that are less likely to have access to multi-disciplinary care, particularly to families who are low income with limited resources such as those served by SafeCare agencies.

A key benefit for the remote delivery of weight management counseling is to increase accessibility to families, especially in more rural areas. However, the family must have the necessary equipment including an internet-enabled device (eg, smartphone, tablet, or computer that is connected to the internet via either a cellular network or WiFi). A recent study of the virtual delivery of SafeCare indicated that many families experienced limited broadband access and technology fatigue, resulting in the need to deliver shorter counseling sessions less than 30 minutes in length [8]. Online interactions may also lessen rapport between the provider and family due to limited ability to see nonverbal cues such as body language. A prior study of a hybrid version of SafeCare, including both face-to-face and virtual sessions, indicated that technology assistance offered efficiencies to the providers in terms of preparation for sessions, but the provider spent more time engaged in rapport-building activities with the family when delivered remotely [22].

Importantly, the parents and children in the pilot study expressed high levels of satisfaction with the remotely delivered program. Children rated satisfaction with talking to their counselor about eating healthier foods as higher than talking with their parent about eating healthier foods, highlighting the effectiveness of remote counseling but the need for further support of the parent-child interaction regarding healthy behavior change. Our findings expand upon a prior study of 360 children and parents randomized to a telehealth family-centered weight management arm in which parents had high levels of engagement and satisfaction with a combination of interactive text messaging and telehealth video calls [23]. A systematic review indicated noninferiority in children’s weight status improvement in telehealth versus in-person treatment delivery, with no difference in attrition rates and consistently high parental satisfaction with telemedicine [24].

Further, these findings add information that SafeCare providers report they are willing and interested in being trained in delivering weight management and believe their families would find this approach with digital tools acceptable. The integration of weight management into a previously existing structured parenting program provides an opportunity for large-scale and rapid dissemination. Families who receive services from SafeCare are often experiencing cumulative risk and have many needs, some of which are not directly related to abuse or neglect. Because SafeCare is broadly disseminated, training providers who already have a connection with these vulnerable families can be a vehicle for delivery of prevention programming that targets other public health issues a family may be experiencing. Should DRIVE prove beneficial, it could be offered as a module of additional services that families could receive. As detailed by the survey of SafeCare providers, there is a perceived need for services such as DRIVE, and SafeCare providers are willing to be trained to provide these services.

The pilot study observed a −0.14 reduction in BMIz (−0.18 among youth who were overweight or had obesity) with 8 counseling sessions delivered over a 13-week period. This reduction is greater than a prior 12-month study that observed −0.09 BMIz among children receiving both enhanced standard of care arm and individualized telehealth coaching (text messages 2x/week and telephone/video sessions every other month) [25] and greater than similar family-based weight management interventions according to a recent Cochrane review of interventions that lasted 6 months or longer [26]. However, the total contact hours did not meet the US Preventive Services Task Force recommendations of at least 26 hours to align with prior efficacious interventions [2], and the BMIz reduction did not meet previously suggested threshold of −0.25 for cardiometabolic improvement [27]. Importantly, in the pilot study, only 4 of the 10 children had obesity and an additional 4 were overweight, and it is not known if these children had cardiometabolic dysregulation. Future work should follow children over a longer time course to determine if BMIz reductions are sustained and accrue longer-term health benefits.

Increasing the dosage of telehealth weight counseling may increase weight loss. For example, a prior study of Kurbo, a commercially available weight management program delivered over a mobile app with video coaching sessions, showed that children who engaged in more telehealth coaching sessions over a longer duration had greater weight loss compared to those with less engagement [28], albeit the level of engagement was self-selected by the family and not randomly assigned. Similarly, a three-arm nonrandomized cohort study observed significantly reduced BMIz among children who opted into a multicomponent technology intervention that included family-based behavioral group treatment, a digital tablet with a fitness tracking app, and
individually tailored telehealth coaching sessions, compared to those who received only the group counseling or the group counseling with fitness app [29]. Programs must strike a balance between families’ compliance/adherence to counseling sessions and expected weight reduction. The convenience of telehealth and digital tools may enable a sufficient amount of engagement that is both effective and acceptable to families.

Limitations
A limitation of these studies is the one-arm design of the pilot feasibility study without a control or comparator condition and the need for further verification in a larger randomized controlled trial. It is possible that BMIz fluctuations were influenced by maturation bias or regression to the mean [30], though the observed effect size was similar to prior pediatric weight management interventions [26]. Another limitation is the use of BMIz to examine change over time, as researchers have identified concerns with z-score for children with a BMI above the 97th percentile [31]. However, only one child in the sample had a BMI exceeding 97th percentile, so it was determined that this metric was appropriate.

Implications for Research and Practice
Our preliminary work demonstrates that DRIVE is an efficacious childhood weight management program capable of being delivered as a module within existing home-based programs, such as SafeCare, and adaptation of DRIVE to include mHealth would benefit both families and SafeCare providers. Families adhered to and were highly satisfied with the telehealth counseling sessions, the wireless scale and weight graph to track child weight, and the pedometer to track child physical activity. These findings are consistent with emerging research documenting that families are responding well to SafeCare delivery via technology, a delivery approach that was implemented as a result of the COVID-19 pandemic [8]. Integrating the intervention into a comprehensive smartphone app or website may enable a more seamless delivery system of both self-monitoring tools and ongoing remote interaction with the counselor. There are many future areas of investigation for mHealth DRIVE, including measuring the effects on weight-related behaviors including dietary intake and physical activity, examining specific feature utilization of the intervention components (such as sharing photos of food preparation with the counselor) and how this relates to the effectiveness of the intervention, and the extent to which the relationship with the counselor drives health outcomes in the family.

Digital tools may present an opportunity for a hybrid approach to blend in-person care with remotely delivered care, bridging the gap between counseling sessions by equipping parents and children with tools to continue their self-monitoring and assist them in implementing the health lessons into their daily lives. SafeCare providers overwhelmingly indicate the perceived need, and willingness to deliver, such a program. Our future work aims to test the feasibility and effectiveness of the mHealth DRIVE program over a longer term to manage children’s weight and improve health-related parenting skills within the context of SafeCare’s telehome visit delivery model. The ultimate goal is to package a turn-key weight management program for families of children with obesity, deployed using mHealth tools for wide-scale dissemination.

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Conflicts of Interest
Georgia State University and Pennington Biomedical Research Center and Louisiana State University have an interest in the intellectual property surrounding the curriculum of the DRIVE intervention, and JRS, JWA, and CKM are inventors of the intervention. Although not the focus of the current paper, Pennington Biomedical Research Center and Louisiana State University own the intellectual property surrounding SmartLoss, SmartIntake, and the Remote Food Photography Method, and an author (CM) is an inventor of the technology.

References


Abbreviations

DRIVE: Developing Relationships that Include Values of Eating and Exercise

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Original Paper

Understanding Parents’ Experiences When Caring for a Child With Functional Constipation: Interpretive Description Study

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Abstract

Background: Pediatric functional constipation (FC) is a common but serious medical condition. Despite significant effects on children, families, and the health care system, the condition is typically undertreated. Parents carry the primary responsibility for complex treatment programs; therefore, understanding their experiences and needs may offer a critical perspective toward improving clinical care.

Objective: The aim of this study is to understand and give voice to parents’ experiences and information needs when caring for a child with FC. The ultimate objective is to build an evidence base suitable for creating a digital knowledge translation tool to better support parents caring for a child with FC.

Methods: This qualitative design used an interpretive description methodology to generate findings aimed at improving clinical care. One-on-one, in-depth interviews were completed either in person or through web-based teleconferencing to explore parents’ perspectives. Data collection and analysis occurred concurrently.

Results: Analysis of 16 interviews generated 4 major themes: *living in the shadows*; *not taken seriously*, with a subtheme of *persevering and advocating*; *missing information and misinformation*; and *self-doubt and strained relationships*. One minor theme of *affirmative influences that foster resilience and hope* was identified.

Conclusions: Parents have unmet needs for support and information related to pediatric FC. To address gaps in current care provision, decision makers may consider interventions for clinicians, resources for parents, and shifting care models to better meet parents’ needs.


KEYWORDS
constipation; child; parents; caregivers; qualitative research

Introduction

Background

Constipation among children is common and often mistaken for a mundane nuisance rather than a serious medical condition. More than 95% of pediatric constipation cases are attributed to functional constipation (FC), which occurs without a particular medical, genetic, anatomic, or physiologic cause. Estimates are that at least 1 in 10 children worldwide is affected by pediatric FC [1,2]. FC can present with severe symptoms such as recurrent abdominal pain, painful defecation, fecal incontinence, urinary incontinence, and urinary infections. Pain, toilet avoidance, and stool withholding behaviors worsen the condition by further perpetuating fear of defecation, causing colonic dilation, and
dampening neural feedback about the need to defecate. Despite being very common, pediatric FC is often underrecognized and undertreated [3]. Without effective treatment, most children develop chronic FC, with symptoms continuing through their adult years [4]. In addition, children and families experience psychological, emotional, and social consequences of FC [5-7]. For example, school attendance and peer relationships are understandably compromised by pain and incontinence. Families also report high levels of stress and decreased quality of life [5-7]. Finally, pediatric FC is a financial burden on families and health care systems [8]. Families face inflated expenses such as medications, laundry, and clothing, in addition to indirect effects such as lost income because of caregiving. Similarly, health care systems are burdened with preventable urgent care visits and high usage rates of specialist services [8,9]. Clinical practice guidelines (CPGs) describe a variety of treatment options [10-14]; however, the bulk of responsibility for implementing, monitoring, and adjusting therapies falls to parents. Certainly, clinicians can provide parents with accurate information about the condition and treatments, but improving care also requires that health care professionals move beyond their own perspective of the condition and acknowledge the unique experiences of families living with a child affected by FC. Specifically, parental experiences critically shape their information and support needs [15]. Therefore, an in-depth understanding of parents’ experiences and self-identified needs when caring for a child with FC is a necessary step to ensure that clinicians can provide relevant education and support.

Although parental education is an important part of treatment for pediatric FC [10-12], there is a lack of research about parental perspectives of pediatric FC. A recent systematic review on the topic included only 13 studies examining parents’ experiences caring for a child with FC [16]. The primary cited limitation of the review was the small number of included studies [16]. Furthermore, there was a predominance of quantitative studies that focused on quality of life measures, which are helpful in substantiating the familial effects of childhood FC but are not optimal in understanding how health care providers can help mitigate negative experiences and outcomes [16]. Suggestions for future research include a more in-depth exploration of how to best meet parents’ information and support needs in light of the dynamic nature of the condition and its profound effects on families [16].

**Objectives**

The initial aim of this study is to understand and give voice to parents’ experiences and information needs when caring for a child with FC. The ultimate objective is to build an evidence base suitable for creating a digital knowledge translation (KT) tool to better support parents caring for a child with FC.

**Methods**

**Design**

The study sought to answer the research question: *What are parents’ experiences and information needs when caring for a child with FC?* Because our ultimate objective was to develop knowledge that could be used to inform and improve clinical practice, we chose the interpretive description (ID) methodology [17] to foster the applicability of our results. ID methodology was developed specifically for practice-oriented sciences, to generate findings aimed at improving clinical care [17], which aligns with our pragmatic philosophical approach for this research project.

**Recruitment**

Potential participants were introduced to the study through social media posts shared on child health and parenting groups (eg, Facebook, Twitter). Physical posters were also displayed in locations frequented by families (sports facilities, libraries, health care waiting rooms, etc) in a medium-sized city in Canada. The posts described the purpose of this study and the desire to speak with the parents of children with FC. In addition, we engaged in snowball sampling by asking participants whether they knew other parents who may be interested in contributing to this study. Recruitment was active from May 2019 until data collection was complete in October 2019.

**Ethical Considerations**

Ethical approval from the relevant research ethics board was granted before the initiation of the study. Each potential participant received an information sheet, which provided details on the purpose of the study, identified the potential risks and benefits, and explained the voluntary nature of their participation. Participants were given an opportunity to ask questions about the research and were free to withhold consent for any reason.

All procedures were in accordance with the ethical standards of the University of Alberta Research Ethics Office (Pro00087548) and the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

**Data Collection Methods**

We used one-on-one, in-depth interviews to explore parents’ experiences when caring for a child with FC. The interviews were completed either in person or through web-based teleconferencing, depending on the participant’s preference and geographic location. The interviewer (AT) had experience conducting qualitative interviews and providing care as a clinician for children with FC. The interviewer did not have any pre-existing personal or professional relationships with the participants. The interviewer spoke with the participants at the beginning of the interview to discuss the reasons for conducting this research (to understand parental experiences and subsequently develop resources for parents) and to share the interviewer’s relevant clinical background—caring for families affected by pediatric FC and noting the challenges they often encountered in managing the condition. The interview style was conversational, and the participants were encouraged to discuss aspects of their experiences they deemed most important. The interviewer also used a semi-structured guide (Multimedia Appendix 1) with open-ended questions. Interview questions were developed based on previous research [18-20] and clinical experience of the team. Prompts and spontaneous questions were used to facilitate participant comfort and collection of high-quality data. Interviews were recorded and transcribed verbatim by a professional transcriptionist. Data were
deidentified (i.e., removal of identifying data such as city names, people names, institution names) to ensure confidentiality.

Sample
Participants were included if their child met diagnostic criteria for pediatric FC (Multimedia Appendix 2) and were willing to discuss their experiences with the interviewer. Screening was conducted by the interviewer as a preamble to the interview to ensure that participants’ stories reflected experiences of childhood FC rather than other conditions. As recruitment was most successful through web-based platforms, participants came from diverse geographical locations across North America.

On the basis of existing literature examining parental perspectives of pediatric FC and methodological recommendations, we anticipated that a sample size between 10 and 20 participants would be adequate to generate clinically significant knowledge [17,21]. The decision to end data collection was an ongoing topic of discussion within the research team and based on the processes of data analysis. Specifically, the occurrence of redundancy within the themes and rich substantiation suggested that data collection could be stopped.

Data Analysis
We followed guidance from the applied methodology of ID [17] throughout data collection and analysis. We conducted data collection and analysis concurrently to promote data immersion as an important step toward a more thorough interpretation of experiences [17]. Interview transcripts were exported into NVivo 12 software to manage the data. Our analytic approach avoided quantification, instead of using thematic and inductive traditions [22,23]. Our analysis followed the processes of engaging with the data, organizing the data, finding patterns within the data, making sense of the patterns, and finally, developing patterns and associations into meaningful findings for applied practice [17]. The process was initiated by the first author, who also conducted the interviews, and then was verified by the author team. Reflexive journaling and field notes were used during data collection and analysis to examine potential bias, build an audit trail, and support rigor.

Rigor
Developers of ID emphasize that the clinical expertise of researchers strengthens the design and rigor of the research [17,24]; therefore, the experiences of clinicians on our research team were seen as a benefit. One member of the research team conducted all the interviews to maintain consistency. The interview guide was reviewed by topic experts and a parent advisory group to enhance credibility and ensure that the questions could elicit meaningful information from participants. A study log was maintained during the research to document and account for methodological decisions. Data were analyzed and findings were collaboratively critiqued by the research team with the intent to develop epistemological integrity, representative credibility, analytic logic, and interpretive authority [17] to ensure high-quality research. Following ID guidance, we did not conduct member checking because of the risks of swaying interpretation and impeding the formation of meaningful clinical implications [17,25]. The study followed the Standards for Reporting Qualitative Research (SRQR) [26] (Multimedia Appendix 3).

Results
Overview
A total of 16 parents of children with FC provided informed consent and participated in this study. Our analysis generated 4 major themes: (1) living in the shadows; (2) not taken seriously, with a subtheme of (i) persevering and advocating; (3) missing information and misinformation; and (4) self-doubt and strained relationships. We identified one minor theme of affirmative influences that foster resilience and hope. The demographic details of the participants are presented in Table 1. All the participants in this study self-identified as caregivers with primary responsibility for managing FC. One of the parents interviewed had more than one child with FC. Participant interviews were randomly assigned a numerical code that was used as a reference marker (e.g., P3) for quotes presented to support the themes in our results.
Table 1. Participant characteristics (N=16).

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Participants, n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Preferred gender identity</strong></td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>16 (100)</td>
</tr>
<tr>
<td><strong>Number of children</strong></td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>4 (25)</td>
</tr>
<tr>
<td>2</td>
<td>8 (50)</td>
</tr>
<tr>
<td>3</td>
<td>2 (13)</td>
</tr>
<tr>
<td>4 or more</td>
<td>2 (13)</td>
</tr>
<tr>
<td><strong>Affected child’s age (years)</strong></td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>1 (6)</td>
</tr>
<tr>
<td>4</td>
<td>4 (25)</td>
</tr>
<tr>
<td>5</td>
<td>4 (25)</td>
</tr>
<tr>
<td>6</td>
<td>5 (31)</td>
</tr>
<tr>
<td>7</td>
<td>0 (0)</td>
</tr>
<tr>
<td>8</td>
<td>0 (0)</td>
</tr>
<tr>
<td>9 or older</td>
<td>2 (13)</td>
</tr>
<tr>
<td><strong>Education level</strong></td>
<td></td>
</tr>
<tr>
<td>High school</td>
<td>1 (6)</td>
</tr>
<tr>
<td>Postsecondary</td>
<td>15 (94)</td>
</tr>
<tr>
<td><strong>Yearly family income in Can $ (Can $1.00=US $0.78)</strong></td>
<td></td>
</tr>
<tr>
<td>&lt;20,000 (15,600)</td>
<td>1 (6)</td>
</tr>
<tr>
<td>20,000-40,000 (15,600–31,200)</td>
<td>1 (6)</td>
</tr>
<tr>
<td>40,000-60,000 (31,200–46,800)</td>
<td>4 (25)</td>
</tr>
<tr>
<td>60,000-80,000 (46,800–62,400)</td>
<td>2 (13)</td>
</tr>
<tr>
<td>&gt;80,000 (62,400)</td>
<td>8 (50)</td>
</tr>
<tr>
<td><strong>Duration of symptoms (years)</strong></td>
<td></td>
</tr>
<tr>
<td>&lt;1</td>
<td>1 (6)</td>
</tr>
<tr>
<td>1-2</td>
<td>3 (19)</td>
</tr>
<tr>
<td>&gt;2</td>
<td>12 (75)</td>
</tr>
<tr>
<td><strong>Number of constipation-related health care visits (total)</strong></td>
<td></td>
</tr>
<tr>
<td>0-5</td>
<td>4 (25)</td>
</tr>
<tr>
<td>6-10</td>
<td>6 (38)</td>
</tr>
<tr>
<td>More than 10</td>
<td>6 (38)</td>
</tr>
</tbody>
</table>

Living in the Shadows

Parents in this study expressed strong feelings of isolation attributed to living with a condition that is considered taboo. Discussing bowel habits and incontinence was thought to be a difficult or inappropriate topic in social circles and within the health care context. For example, when parents themselves were open to the conversation, most had experienced or anticipated negative reactions from others. One parent related her sense of isolation, “Nobody talks about it…. So, you feel alone… And nobody wants to talk about poop” (P3). Similarly, another parent explained:

*I think, that for myself…because I don’t know a lot of other parents that are – I don’t know if people just don’t talk about it, so I don’t know how common it is.* [P4]

To combat feelings of isolation, parents typically searched for resources without success to meet their social support needs. Parents were surprised about the lack of discussion groups because many described how it seems there is an online forum for almost every rare disease or condition:

*Something…so you’re not alone, right. Because that’s the thing and you don’t understand why your kid is having so many problems. It’s like somebody or*
something that explains like oh my kids have this issue, so you don’t feel like you’re the only one...Just something you can go to whether it’s like a chat group or a parent group or something. [P5]

Another parent described how she would change things to improve other families’ experiences with pediatric FC:

You know, I think it’s one of those things that people could really benefit from a support group because it’s something that’s so like people don’t wanna talk about, they’re embarrassed about it. [P9]

Another parent simply expressed, “I just feel like we were very much left on our own” (P14).

Not Taken Seriously

Parents shared stories of encounters with health care professionals who did not take their concerns about constipation seriously. In some cases, parents were explicitly told that the symptoms were nothing to be concerned about, and in other cases, parents were implicitly given the impression that they were overreacting. One parent shared her care provider’s dismissive response to her child’s symptoms:

I was always told it would pass, it would pass. Probably listen to the patient a little bit better because they know their body, right, and I – me living with her, I know what’s going on with her. So, listen a little bit closer and maybe have better options than prune juice. [P13]

Similarly, another parent said:

I wish I had been taken seriously right away. You know, not just like she’ll grow out of it, she’ll grow out of it. It’s normal, she’ll grow out of it. It’s like this wasn’t. I don’t know if it ever was. [P9]

One shared the widespread effects of her child’s FC and the trivializing response:

I get that pediatricians are really busy with other things that are, you know, more important than constipation, but like now that he’s in school, it’s affecting his whole class. It’s affecting his teacher. It’s affecting him and his friends. Like it affects a lot of things and it affects us daily. It takes up our time as parents and his time away from his activities and the only real thing that we hear is, oh don’t worry, it’ll end soon. Like how? [14]

One parent reflected on her desire for health care providers to change:

I guess I wish they would learn – they would take it a bit more seriously and understand how it impacts lives and how it impacts – I mean children’s lives. [P7]

Parallel to instances of health care providers not taking the condition seriously, parents themselves described periods of questioning the legitimacy or validity of their own concerns. For example, one parent shared:

I think we could have maybe helped him a lot sooner if I wasn’t so scared to start the Lax-A-Day but I also didn’t want to make an appointment, take someone else’s doctor time...I hate wasting doctors time on what I consider a silly thing...I know it’s not the right way to think of it but like to my point, it had to be urgent enough. [P3]

Similarly, another parent said, “You’re like, oh is that normal or not normal and you kind of doubt yourself” (P2).

Persevering and Advocating

As a result of symptoms and concerns not being taken seriously, parents demonstrated perseverance and became stronger advocates for their child’s health. One parent described her feelings about health care encounters:

I had talked to my doctor about it. Like our doctor and the doctor said like, oh you know, she’s still really young. She’ll grow out of it, all that kind of stuff...eventually after lots of kind of like advocating, I ended up – I was like I need another opinion on this. [P9]

Similarly, another parent stated:

We found that we’ve gone to the doctor a couple of times now and they haven’t been super helpful...and then we wound up back at the doctor because we’re still – she’s still having accidents. [P16]

Parents returned to health care providers repeatedly and asked for referrals to other providers because their child’s condition was worsening without adequate treatment. For example, “I’d asked many times for her to be seen by somebody else just because I need this figured out” (P13). Parental frustration frequently became the catalyst for advocacy. One parent expressed:

They don’t take it serious enough...it would just be nice if there was a doctor that would take you a little more serious. I know lots of kids have it and I get that, but when they get to be older and it’s a school issue, I think like we push. I think we asked – my doctor was out of town so we asked the stand in and then we asked the walk-in clinic and then we asked my doctor. [P5]

Missing Information and Misinformation

Parents caring for a child with FC frequently have unanswered questions about the condition, causes, symptoms, prognosis, and treatment. One parent said:

Maybe I wouldn’t have been so upset about it or, you know, it wouldn’t have been such an overly concern for me if I’d had a little bit more information. [P2]

Similarly, another parent explained the lack of teaching provided about pediatric FC:

I’m saying like you go into the doctor and you’re like this is an issue and they don’t give you...like there’s nothing, they give you nothing. My doctor was just very much like, oh it’s super common and...like not giving you any further advice or resources. [P6]

Parents frequently questioned whether there was an underlying medical cause for constipation. For example, one parent stated:
Maybe something else medically. Like maybe she’s lactose intolerant – we thought well maybe there’s some issues with milk or dairy which, of course, would not be constipation...but we were convinced it was something she was eating. Maybe it was gluten, maybe it was this, maybe it was that. [P1]

Episodes of incontinence often cause parents to question the underlying reason. One parent wondered, “I don’t know if it’s medical or constipation or is it just laziness?” (P5). Similarly, another parent stated:

We had no idea whether she actually like did she have control, did she not have control. Could she feel it, could she not feel it? Was she just ignoring it? Did she need to pay more attention? Like all of these huge question marks. [P9]

Questions about the treatment for pediatric FC were also common. A parent shared concerns about medication use:

You read the Lax-A-Day thing it says, “Adults only”, blah, blah, blah. So, I’m like ‘Are you sure?’ Like it feels wrong...But then, again we’re trying to cut back now on the Lax-A-Day because you can’t be on Lax-A-Day forever, can he? Like I don’t know. [P3]

In addition to having questions about pediatric FC, parents shared instances of misinformation that was detrimental to their child’s care. As explored above in the theme of not being taken seriously, parents were often incorrectly told that the condition would resolve on its own. One parent shared the common false reassurances she received:

It was very much like, no, no, no, he’s fine. And it’s just constipation and he’ll grow out of it and like I feel like everybody I talked to said, he’ll grow out of it. He’ll grow out of it. He’ll grow out of it. And now, two years later, he’s not growing out of it. [P14]

Parents were also commonly given misinformation about dietary changes as treatment. “We were just told to increase fibre, increase water, skip the junk food, but we eat all whole foods anyways” (P4). Similarly, another parent shared, “The doctor said, it’ll get better. You know, just make sure she’s eating healthy, which she does, and it’ll get better. It’ll get better” (P16). Dietary misinformation was problematic because it was ineffective, difficult for families to manage, and delayed further treatment:

The nurse said don’t give her any dairy. And so, we were off dairy for a while and then we were off wheat for a while and it was just like a – none, none of that seemed to make much difference. [P9]

Similarly, another parent reported, “Cut [cheese] out and try to increase the fruits, the vegetables, take away the bread. It was like a constant diet struggle” (P3).

Within this theme, there was one divergent case of a parent who conveyed confidence and felt that they had adequate knowledge about caregiving for a child with FC. The case had minimal health care encounters because the parent felt further support or intervention was not required. Unfortunately, the parent’s knowledge was inferred from personal experience with medical care of an unrelated population and condition, which does not align with current evidence for pediatric FC. Thus, although the participant expressed a divergent view of her experience, the data further substantiated the theme of missing information and misinformation.

Self-Doubt and Strained Relationships

Perhaps the most resounding theme from parents’ stories was the overarching sense of frustration that developed while caring for a child with FC. One parent shared the emotional fragility that pediatric FC had created for her as a parent:

It’s pretty terrible actually. Like I should know how to deal with this. I’m a nurse. Like I was a pediatric nurse. (crying). I should know and everything that I’ve tried didn’t work and I didn’t have any guidance or any help. Like I called the doctor, well it’s you know, the pediatrician – it’s six months to get into her, so I, you know. I’m just trying things on my own. I’m googling how do you deal with this and you know, information and none of it is working and it makes me feel like – I don’t know. Like I should know how to do this, and I don’t. [P14]

Self-doubt and conflict were strongly tied to the previous themes of living in the shadows, not being taken seriously, and missing information and misinformation. One parent clearly expressed the situation stating:

It was just like extremely frustrating because I felt like I wasn’t getting – I wasn’t getting enough support or information from the medical – like the health professionals we were dealing with...Like it’s so frustrating. I’m like if this is so common, why does no one have answers? – it’s just so, so frustrating. [P9]

Symptoms and physiology of pediatric FC were further sources of emotional turmoil for parents:

We are very frustrated and, again, the accidents, I don’t know if it’s because of this issue or because she’s lazy or like because she’s so constipated...it’s the accidents that are driving us crazy. [P5]

Another parent explained:

We’ll tell him fifteen times to go to the bathroom and he won’t and then he’ll have an accident and you feel like – you just get to your boiling point sometimes and you don’t want to yell and get angry, but sometimes you do. [P14]

Finally, relationships frequently became strained as a result of pediatric FC:

It impacts a whole family dynamic, you know. Like our world, it seems like I mean this might sound dramatic, but our world has literally revolved around her bathroom habits for the last three years. [P16]

Another parent expressed the strain related to behavioral interventions, “Like it’s always a fight to get her on the toilet” (P6). Another parent stated, “There’s been lots of fights. Lots of fights. Lots of I hate yous” (P10). Emotional burden related...
to pediatric FC also sparked conflict between parents and eroded parental self-efficacy:

We’re both feeling – neither one of us are confident in our parenting. So, we’re frustrated, and we can argue about it, for sure...I really felt like a failure as a mom. (pause). I don’t know and I still don’t know what to do. I don’t feel like we’re making progress and I don’t feel like I have the confidence to fix it. And then I feel like that kind of – permeates, I guess, into our whole situation. Like if I can’t figure out constipation, how can I figure out big things? [P14]

**Affirmative Influences Foster Resilience and Hope**

Despite the predominantly despondent themes that were reflected in parents’ stories, there were small but significant moments of affirmation that helped bolster parents’ confidence. This is a minor theme of our analysis because the occurrence of positive encounters and resources was unfortunately infrequent. After episodes of misinformation, accurate and understandable explanations of the condition and symptoms were critically important for parents:

They explained the encopresis is like the fact that like you know, when she did get constipated, the accidents would just be like the new poop coming around the old stuff that’s not coming out...it’s just like your muscles are just weak because like they’ve been holding it for so long. Yeah, and I was just like – at first, it just kinda blew my mind and I’m like, why the hell has no one told me about this? [P9]

Validation came from a variety of sources and was always highlighted as an important event within the caregiving experience. For example, one parent found support through the school system:

And it was really just brushed off and it’s still being brushed off until like finally – now that he’s taking up so much time from his teacher, the principal has become involved and she has been our only real advocate and our only – like the principal of the school. Like she’s not a health care provider. You know, like she’s the only person that has really like tried to help at all. [P14]

Parents identified encounters that met their support and informational needs as turning points that rekindled hope and buoyed their confidence. Unfortunately, affirmative influences were meaningful but scarce in parents’ experiences. Specifically, many parents did not relate any positive encounters or support at all throughout their caregiving journey. One parent explained:

I told them this has been an ongoing issue. This isn’t getting any better. This isn’t an issue we’ve had for six months. This is an issue we’ve had for over three years now. [P16]

**Discussion**

### Principal Findings

Findings from our exploration of parents’ experiences with pediatric FC parallel and expand upon results from previous research in the field. In a 2003 study, researchers examined parents’ health care encounters related to childhood constipation and found similar themes of “dismissed and fobbed off, asserting the need for action, and validation and acknowledgment” [21]. The continuity of these findings with ours suggests that parents’ perceptions of encounters with health care providers related to pediatric FC have not improved significantly over the last 17 years. Despite the widespread prevalence of the condition [1,2] and advances in understanding childhood FC [3,4], parents’ concerns continue to be minimized and clinicians’ treatment discussions lag behind or are incongruent with symptom severity. In other words, when health care providers acknowledge that pediatric FC requires treatment (which in itself may occur belatedly, if at all), the level of intervention is often inadequate for the advanced nature of symptoms described by parents.

Similar to exploring patient and family experiences, measuring quality of life is considered an important way to understand the effects of a health condition or treatment on “patients’ lives, rather than just on their bodies” [27]. Numerous studies have highlighted the diminished quality of life of parents and families living with pediatric FC [28-32]. For example, 3 studies found that increased family conflict, impaired family functioning, and increased parental worry or stress were related to the presence of fecal incontinence [29,30,32]. Furthermore, Wang et al [31] found that the caregivers of children with FC gave lower ratings of their daily activities and family relationships, in addition to reporting lower physical, emotional, social, cognitive, and communication scores compared with those of the caregivers and families with healthy children. Although quality of life data provide a broad assessment of the effects of a health condition and are a central contribution to the field, qualitative methods are helpful in adding important context by exploring why and how families are affected. In this study, parental perspectives provide insights into the significant physical, emotional, and psychological burden on caregivers. Parents’ feelings of isolation and frustration were related to incontinence and further compounded by nonsupportive interactions and misinformation. Parents’ experiences of being told erroneously that pediatric FC would resolve, feeling blamed for the condition or lack of treatment success, and struggling to talk about the condition may help explain the widespread and profound impairments in quality of life for families affected by pediatric FC [28-32].

A 2019 study examining the prevalence of defecation disorders in children concluded that childhood constipation is likely underestimated by parents who may not consider symptoms sufficient to be labeled a medical condition [2]. The findings seem to be in contrast to our data, which found that parents were more frequently dismissed by health care providers rather than they being dismissive of the child’s symptoms. One potential explanation for this difference could be the relative disease severity of the parents surveyed in the 2 studies. Specifically,
the cross-sectional study included a random selection of parents from the general population and was, therefore, more likely to include parents with early or mild manifestations compared with parents included in this study whose children all met full diagnostic criteria for pediatric FC. The findings from this study offer a relevant counterpoint, meaning that although parents and families may underestimate early symptoms, once the magnitude of the condition becomes evident, health care providers may be more of a barrier to recognition and diagnosis than parents.

**Clinical Implications**

Our exploration of parents’ experiences of caring for a child with FC provides important insights toward improving clinical care for this difficult condition. CPGs, which are intended to support clinicians and optimize care, identify family education about pediatric FC as a key component of treatment [10-12]. Unfortunately, our results suggest that this step is commonly missing in health care encounters and that some providers even contribute to misinformation. As our data were focused on parental perspectives, we cannot report the reasons for CPG deviations. Given the time-consuming nature of consultations to provide emotional support and education, it is possible that care providers may be tempted to defer, rush through, or simply struggle to fit these practices into already-busy schedules. On the basis of parents’ reluctance to initiate discussions about bowel concerns, it may be prudent for professionals to recognize that effects may be more severe and have persisted for a significant duration by the time these issues are brought to their attention. In contrast to the temptation to offer hasty reassurance, clinicians may need to reframe their thinking toward acknowledgment, education, and active treatment. For example, explaining that the condition is common can be a method of validating parents’ concerns and mitigating parental feelings of guilt but should not be conflated by suggesting that the symptoms are normal or do not require treatment. Improving the quality of health care encounters may require education or interventions to improve the responses and treatment knowledge of health care providers. Similar to findings from a previous study about medication adherence [33], parents commonly expressed a lack of information about medication use; therefore, discussions about dosing, duration of use, side effects, and safety are likely to be well received by parents. Finally, clinicians should be attuned to inquiring about parental experiences of isolation and lack of social support during assessment and include these factors as part of treatment plans [10-12]. In addition to the existing system constraints that disincentivize lengthy consultations, it is unlikely that specialty care providers or primary care clinicians alone can adequately meet complex parental needs. Consideration of alternative care models, such as integration of nursing and allied health members, may be helpful to more accurately and consistently meet parents’ support needs when caring for a child with FC [34-36].

**Future Steps**

The results of this study are an important foundation for creating resources that directly address parents’ experiences and self-identified needs when caring for a child with FC. Developing support such as digital KT tools that target parents’ information needs may improve families’ experiences of living with pediatric FC. For example, parents seek answers to concrete questions about medication dosing, titration, side effects, safety, and long-term use. Sharing information with parents about digestive physiology, including how constipation can contribute to fecal incontinence, may be helpful in empowering parents’ caregiving when faced with the uncertainty and frustration that arise from a child’s stool accidents. In addition, the emotional toll of pediatric FC on families was often underacknowledged, wherein parents’ caregiving abilities were hindered because of self-doubt and guilt. Creating resources that validate parental concerns and experiences can be an important contribution to meeting the support needs of parents caring for a child with FC. Finally, in light of our findings related to health care providers, future research exploring health care professionals’ knowledge of pediatric FC and their experiences working with affected families can clarify the challenges and barriers to improving care provision for this condition.

**Limitations**

Although the recruitment was open to all parents, we only received interest from mothers. The interviewer asked whether any other caregivers from each family would be interested in sharing their perspective; however, we did not successfully recruit any further participants. Therefore, our results may not reflect the experiences of fathers and nonprimary caregivers. Parents who shared their story for this study were typically from higher education and income levels; therefore, experiences of parents with lower levels of education or income may not be adequately captured in our findings. In addition, the sample may reflect bias because of the self-selection nature of the recruitment process.

**Conclusions**

Understanding parents’ experiences when caring for a child with FC is an important and often overlooked step toward improving care for this difficult condition. Our findings indicate that parents have significant unmet needs for support and information related to pediatric FC. To address gaps in current care provision, decision makers may consider interventions for clinicians, resources for parents, and shifting care models to better meet parents’ needs.
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Authors’ Contributions

AT, SM, EW, and SS conceived and designed this study. AT was responsible for data acquisition. All authors contributed to the analysis of the data. AT drafted the preliminary version of the manuscript. SM, EW, and SS critically revised the work for important intellectual content. All authors approved the final manuscript and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. SS obtained the research funds through which this research was conducted.

Conflicts of Interest

None declared.

Multimedia Appendix 1

Draft interview guide.

[PDF File (Adobe PDF File), 76 KB - pediatrics_v4i1e24851_app1.pdf]

Multimedia Appendix 2

ROME IV diagnostic criteria for pediatric functional constipation.

[PDF File (Adobe PDF File), 14 KB - pediatrics_v4i1e24851_app2.pdf]

Multimedia Appendix 3

Standards for Reporting Qualitative Research (SRQR) checklist.

[PDF File (Adobe PDF File), 158 KB - pediatrics_v4i1e24851_app3.pdf]

References


Abbreviations
- CPG: clinical practice guidelines
- FC: functional constipation
- ID: interpretive description
- KT: knowledge translation

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Parents’ and Students’ Perceptions of Telepractice Services for Speech-Language Therapy During the COVID-19 Pandemic: Survey Study

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Abstract

Background: The ongoing COVID-19 pandemic has resulted in the suspension of face-to-face classes and a considerable increase in the use of telepractice services in speech-language pathology. However, little is known about parents’ and students’ satisfaction with telepractice services and their preferences for different service delivery modes. These factors may affect therapy effectiveness and the future adoption of telepractice.

Objective: We evaluated students’ and parents’ perceptions of telepractice efficacy and their preferences for different service delivery modes (ie, on-site practice vs telepractice). We also identified factors that affect parents’ and students’ preferences for different service delivery modes during the COVID-19 pandemic.

Methods: A 19-question survey on telepractice satisfaction and preferences was administered to 41 Hong Kong Chinese students and 85 parents who received telepractice services from school-based speech-language pathologists during the COVID-19 class suspension period. In addition to providing demographic information and data on the implementation of telepractice services, all participants were asked to rate their perceptions of the efficacy of telepractice services and compare on-site practices to telepractice on a 5-point Likert scale (ie, 1=strongly disagree/prefer the use of on-site speech-language therapy services and 5=strongly agree/prefer the use of telepractice services).

Results: Despite the fact that telepractice efficacy was highly rated by parents (95% CI 3.30-3.66) and students (95% CI 3.21-3.76), both groups believed that telepractice was less effective than on-site practices (parents: 95% CI 2.14-2.52; students: 95% CI 2.08-2.65). Moreover, parents preferred on-site practices over telepractice (95% CI 2.04-2.43), whereas students did not prefer one mode of practice over the other (95% CI 2.74-3.41). A significant association between telepractice efficacy and a preference for telepractice services was found only among the students (τ=.43, P<.001), not the parents (τ=.07; P=.44).

Conclusions: Although telepractice is an acceptable alternative service delivery option for providing speech and language therapy services to school-aged individuals, speech-language therapists and parents must play a more proactive role in telepractice services to facilitate effective communication between clinicians and parents.


KEYWORDS
eHealth; telepractice; speech and language pathology; user satisfaction; COVID-19; school-based service

Introduction

As of January 2021, over 90 million people have been infected with the SARS-CoV-2 virus. This has necessitated social distancing and school closures worldwide. As a result, telehealth (ie, the use of audio or videoconferencing technology to provide health care services) has received increasing attention. Telehealth care has been regarded as an alternative to
face-to-face care in many countries [1,2]. Furthermore, speech-language pathologists have engaged in telepractice over the past 2 decades in various countries [3-6]. The efficacy of telepractice has been supported by scientific research on speech, language, voice, and fluency disorders across different age groups [7-9]. Additionally, telepractice has been deemed valid and effective by different professional organizations [10,11]. With the COVID-19 pandemic seriously disrupting the provision of speech and language therapy services, telepractice services have been increasingly adopted and regarded as the best option for delivering speech and language therapy during the pandemic [12,13].

Despite the increasing adoption of telepractice in schools, various stakeholders have held different beliefs about telepractice. Although several surveys have shown that school-based speech-language pathologists doubt the efficacy of telepractice, others have revealed a positive attitude after using telepractice services [12,14,15]. However, parents’ and children’s perceptions of telepractice are not well understood. A few studies have examined parents’ and students’ satisfaction with telepractice programs, but the findings have been mixed. In a pilot survey, 13 teachers and 8 parents from a remote school were highly satisfied with the progress brought about by telepractice [8]. Positive findings were also noted in parents’ and students’ responses to a survey on web-based speech and language interventions that were conducted by university clinics [8,16]. In contrast, an interview study of 5 parents raised concerns about poor telepractice engagement by students and ineffective communication between parents and clinicians in telepractice services [17]. These factors may lower people’s acceptance of school-based telepractice services [17]. Given the high rate of telepractice adoption in school settings during the pandemic [12,13], a survey study on parents’ and students’ satisfaction with telepractice could reveal the perceived efficacy of these services.

Perceived efficacy is an important measure in speech and language therapy for both on-site practices and telepractice, because it reflects the effectiveness of the therapy and students’ and parents’ motivations for undergoing the therapy [18,19]. The Davis’ Technology Acceptance Model also argues that perceived efficacy, which is based on perceived usefulness and convenience, influences the future adoption of technology [20]. Perceived efficacy can be reflected by people’s engagement with therapy sessions, which correlates with children’s treatment outcomes [21]. Moreover, the amount of therapeutic skills that families practice during their daily routine and the collaboration between clinicians and parents affect the generalization of treatment [22]. Therefore, investigating parents’ and students’ perceptions of telepractice efficacy and their involvement with telepractice and daily therapeutic practices are critical for evaluating treatment fidelity.

Previous studies have largely focused on students’ and parents’ satisfaction with research-oriented telepractice, but none have investigated clients’ and parents’ preferences for different modes of practice. Since service delivery modes have expanded during the pandemic, students’ and parents’ preferences for different delivery models are critical for designing a future service delivery model for school-based speech and language therapy services. Thus, in this study, we examined how clients’ therapy characteristics, including age, comorbidity, and parent support, influence their preferences for different modes of service. This information may inform speech-language pathologists about selecting appropriate students for telepractice services [10].

In summary, the following 3 research questions were addressed in this satisfaction survey study: (1) what are parents’ and students’ perceptions of telepractice efficacy; (2) do parents and students prefer on-site practices or telepractice; and (3) what are the critical factors that affect parents’ and students’ preferences for different service delivery modes?

**Methods**

**Survey Design and Development**

**Survey Summary**

We developed a web-based survey for both parents and students to evaluate school-based speech and language therapy practices in Hong Kong (see Multimedia Appendix 1). To meet internal clarity, construct, and content validity criteria, all survey questions were independently reviewed by 3 school-based speech-language pathologists. This review ensured that the survey’s wording, content, and question order were clear and appropriate. The survey questions were revised and finalized in accordance with the speech-language pathologists’ suggestions. All respondents completed the survey in about 10 minutes. Ethics approval was granted by the Human Research Ethics Committee of University of Hong Kong, and participants signed consent forms before completing the survey.

The survey for parents and students consisted of 4 sections, including (1) the implementation of telepractice, which consisted of 2 items; (2) telepractice efficacy, which consisted of 7 items for parents and 4 items for students; (3) the comparison between telepractice and on-site practice, which consisted of 6 items for parents and 5 items for students; and (4) demographics, which consisted of 5 items. All responses for sections 2 and 3 were based on Likert-type scale scores, which ranged from 1 (ie, strongly disagree) to 5 (ie, strongly agree).

**Section 1: Implementation of Telepractice**

The 2 items in this section assessed the amount of therapy students received and how frequently students used telepractice services during the COVID-19 class suspension period.

**Section 2: Telepractice Efficacy**

The 7-item survey for parents included questions about whether telepractice was effective in enhancing their child’s language skills, meeting their child’s needs, engaging with their child, and providing satisfaction with the amount of therapy their child received (Cronbach α=.94). The 4-item survey for students included questions about whether telepractice services met their needs and whether they enjoyed telepractice services (Cronbach α=.84).

**Section 3: Comparison Between Telepractice and On-site Practice**

The 6-item parent survey included questions about whether telepractice services for speech therapy provided better...
communication than on-site speech and language therapy. There were also questions regarding the implementation of home therapy practices (Cronbach $\alpha=.89$). The 5-item student survey included questions about whether students learned better language skills and exhibited better engagement with on-site practices than with telepractice (Cronbach $\alpha=.88$).

**Section 4: Demographics**

The 4 items in this section were used to collect information on each student’s grade, gender, special education needs status, and family income.

**Participants**

From July to August 2020, 85 parents (ie, 75 mothers and 10 fathers) and 41 students (ie, 7 girls and 34 boys) participated in our web-based survey. Based on the last 4 digits of participants' telephone numbers, 27 families participated in both the parent and student surveys. These 27 families accounted for the 31% (27/85) of parents and 65% (27/41) of students who participated. The families who responded to both the parent and student questionnaires represented students from Grades 1-7 (parents’ questionnaire: median=Grade 3; students’ questionnaire: median=Grade 4). In terms of students' comorbidities in the parent survey, the most prevalent special educational needs subtype was autism spectrum disorder (53/85, 62%), followed by attention deficit/hyperactivity disorder (33/85, 38%), specific learning difficulties (20/85, 23%), intellectual disabilities (3/85, 3%), hearing impairment (2/85, 2%), visual impairment (1/85, 1%), and physical disabilities (1/85, 1%). Additionally, 12% (11/85) of students had no comorbidities except for speech and language disorders. In terms of students' comorbidities in the student survey, the most prevalent special educational needs subtype was autism spectrum disorder (24/85, 58%), followed by attention deficit/hyperactivity disorder (15/41, 36%), specific learning disorders (6/41, 14%), intellectual disabilities (1/41, 2%), and visual impairment (1/41, 2%). Additionally, 21% (9/41) of students had no comorbidities except for speech and language disorders. Around half of the participants (parents’ survey: 42/85, 49%; students’ survey: 22/41, 53%) had an average monthly family income that fell below the median for average household income (ie, around US $3290).

To achieve a Cronbach $\alpha$ value of .05 and a moderate effect size (ie, Cohen $d=0.5$), a statistical power of .99 and .86 was needed for 85 parents and 41 students, respectively. This was determined by using G*Power 3 software (G*Power Team) [23]. In addition, good quality results can be obtained by performing a factor analysis on samples with at least 50 people or samples with a factor loading value of $>.60$ [24].

**Results**

**Implementation of Telepractice**

Most students reported that they had fewer than 5 telepractice sessions during the pandemic (parent’s survey: 73/85, 85%; students’ survey: 31/41, 75%). In terms of session frequency, the most common amount of therapy was 1 session per month (parents’ survey: 35/85, 41%; 36%; students’ survey: 15/41, 36%), followed by 1 session per 2 weeks (parents’ survey: 25/85, 29%; students’ survey: 15/41, 37%), and 1 session per week (parents’ survey: 21/85, 24%; students’ survey: 12/41, 29%).

**Telepractice Efficacy**

Parents and students had positive views of the efficacy of telepractice with respect to their understanding of the treatment goals (parents: mean 3.48, SD 0.84; 95% CI 3.30-3.66; students: mean 3.49, SD 0.87; 95% CI 3.21-3.76) and the ability of telepractice services to meet the needs of students (parents: mean 3.24, SD 1.03; 95% CI 3.01-3.46; students: mean 3.49, SD 0.84, 95% CI 3.22-3.75). Based on the parents’ responses, parents had positive views of students' enjoyment of telepractice services (mean 3.29, SD 1.14; 95% CI 3.05-3.54) and the ability of telepractice services to enhance students’ language abilities (mean 3.33, SD 1.01; 95% CI 3.11-3.55). Based on the students’ responses, students had a neutral view of telepractice efficacy with regard to (1) enjoyment (mean 3.32, SD 1.08; 95% CI 2.98-3.66) and (2) language ability enhancement (mean 3.29, SD 0.96; 95% CI 2.99-3.59). Independent 2-tailed sample $t$ tests revealed that there were no significant differences in the above views between parents and students (enjoyment: $P=.92$; understanding of treatment goals: $P=.97$; meeting students’ needs: $P=.18$; language ability enhancement: $P=.85$). In addition, parents held a positive view of the progress that students made during telepractice services (mean 3.35, SD 0.96; 95% CI 3.15-3.56) and a neutral view of the amount of therapy that students received (frequency: mean 2.99, SD 1.04; 95% CI 2.76-3.21; amount of therapy: mean 3.21, SD 1.03; 95% CI 2.99-3.43).

**Factors That Affected Telepractice Efficacy**

Our Spearman rank-order correlation analysis showed that there were no significant correlations between student grade and perceived telepractice efficacy (parents: $p=0.03$; $P=.76$; students: $p=0.07$; $P=.65$). The Bayes factor (BF) was computed to evaluate whether the evidence supported the null hypothesis over the alternative hypothesis. BF$_{01}$ values of $>3$ and $>10$ indicated moderate and strong support, respectively, for the null hypothesis [25]. Strong evidence that supported the null hypothesis (ie, no correlation between grade and telepractice efficacy) was found in the parent group (BF$_{01}$=11.34), whereas moderate evidence that supported the null hypothesis was found in the student group (BF$_{01}$=7.84).

**Comparison Between Telepractice and On-site Practice**

Students’ enjoyment of telepractice services and on-site services was comparable, based on the students’ responses (mean 2.93, SD 1.06; 95% CI 2.59-3.26). However, students’ enjoyment of telepractice services was lower in the parents’ responses (mean 2.76, SD 1.02; 95% CI 2.54-2.98). Furthermore, telepractice was rated lower than on-site practice in terms of treatment effectiveness. The aspects of treatment effectiveness included the acquisition of speech and language skills (parents: mean 2.47, SD 0.92; 95% CI 2.27-2.67; students: mean 2.46, SD 0.93; 95% CI 2.17-2.76), communication with speech-language pathologists (parents: mean 2.52, SD 0.88; 95% CI 2.33-2.71; students: mean 2.32, SD 0.82; 95% CI 2.06-2.58), and treatment efficacy (parents: mean 2.33, SD 1.89; 95% CI 2.14-2.52; students: mean 2.37, SD 0.92; 95% CI 2.08-2.65). An
independent 2-tailed sample $t$ test revealed no significant differences in these aspects between parents and students (enjoyment: $P=0.41$; acquisition of speech and language skills: $P=0.97$; communication with speech-language pathologists: $P=0.22$; treatment efficacy: $P=0.83$). In addition, parents rated telepractice lower than on-site practice, in terms of the implementation of therapy practices at home via telepractice services or on-site services (mean 2.46; 95% CI 2.27-2.65).

Parents had a significant negative view of telepractice, with regard to whether they preferred telepractice over on-site practice (mean 2.24; 95% CI 2.04-2.43), whereas students had a neutral view (mean 3.07; 95% CI 2.74-3.41). An independent 2-tailed sample $t$ test revealed a significant difference in preferences for telepractice and on-site practice between parents and students ($t_{124}=4.59; P<.001; d=0.87; 95\%$ CI 0.48-1.26).

Factors That Affected Preferences for Telepractice and On-site Practice

**Grade**

Our Spearman rank-order correlation analysis showed no significant correlations between student grade and participants’

<table>
<thead>
<tr>
<th>Item</th>
<th>Parents $^a$, factor loading value</th>
<th>Students $^b$, factor loading value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Student enjoyment</td>
<td>.857</td>
<td>.552</td>
</tr>
<tr>
<td>Understanding of treatment goals</td>
<td>.798</td>
<td>.941</td>
</tr>
<tr>
<td>Meeting the needs of students</td>
<td>.926</td>
<td>.776</td>
</tr>
<tr>
<td>Enhancing speech and language abilities</td>
<td>.903</td>
<td>.819</td>
</tr>
<tr>
<td>Understanding treatment progress</td>
<td>.914</td>
<td>N/A $^c$</td>
</tr>
<tr>
<td>Appropriate session frequency</td>
<td>.726</td>
<td>N/A</td>
</tr>
<tr>
<td>Appropriate session duration</td>
<td>.670</td>
<td>N/A</td>
</tr>
</tbody>
</table>

$^a$The factor score for the parent group accounted for 73% of the variance in the items. Each item had an eigenvalue of 5.13.

$^b$The factor score for the student group accounted for 69% of the variance in the items. Each item had an eigenvalue of 2.79.

$^c$N/A: not applicable. These items only appeared in the parent questionnaire.

The Kendall rank correlation coefficient, $\tau$, was computed based on the factor scores for telepractice efficacy and preferences for the mode of practice. No significant correlation was found in the parent group ($\tau=.07; P=.44$); the BF for this correlation (BF$_{01}=8.53$) moderately supported the null hypothesis (ie, there is no correlation between telepractice efficacy and preferences for the mode of practice). A significant correlation between telepractice efficacy and preferences for the mode of practice was found in the student group ($\tau=.43; P<.001$).

**Discussion**

**Principal Findings**

Unlike previous telepractice studies, which have largely focused on clinicians’ attitudes, our study examined parents’ and students’ perceptions of telepractice efficacy and their attitudes toward telepractice during the COVID-19 pandemic. We found that students and parents were satisfied with the efficacy of treatments that were provided through telepractice services. Although students and parents had similar preferences for telepractice and on-site practice, parents preferred on-site practices. These findings are discussed in terms of telepractice efficacy and factors that affect engagement with telepractice services.

**Treatment Efficacy**

To examine the relationship between treatment efficacy and preferences for the 2 service delivery modes, we created a composite score based on the factor scores that were obtained from our exploratory factor analysis, by performing principal axis factoring extraction. As shown in Table 1, we obtained a factor score that accounted for 73% and 69% of the variance in the parent and student groups, respectively. All factor loadings were greater than .55.
school-aged students with various disorders [7,28,29]. This evidence suggests that students with special education needs can benefit from treatments that are provided through telepractice services.

**Preference for Telepractice and On-site Practice**

Despite students’ and parents’ satisfaction with telepractice efficacy, students did not prefer one mode of practice over the other, whereas parents preferred on-site practice over telepractice. However, there was no significant correlation between telepractice efficacy and parents’ preference for on-site practice (P=.44). This indicates that other concerns may have influenced parents’ preferences. Interestingly, compared to parents’ views of on-site practice, parents expressed a negative view of telepractice in terms of treatment effectiveness, the implementation of therapy practices at home, and communication with speech-language pathologists. This negative opinion can be explained by the lack of effective communication in telehealth. Due to the lack of personal interaction that occurs in telehealth services, extra communication and visual features for communication are needed to build a rapport between clinicians and parents [29].

For example, when discussing sensitive topics (e.g., diagnosis, comorbidity, and prognosis) on web-based platforms, parents may feel a sense of depersonalization [29,30]. In addition, face-to-face communication has been indicated as a preferred mode of communication in various studies, as face-to-face communication allows for the better observation of visual cues, such as facial expressions and body language [31-33]. Another explanation for parents preferring on-site practices over telepractice is that parents need to provide extra effort and input in telepractice services. In telepractice sessions, parents need to solve technological problems and control students’ behaviors throughout the session. Therefore, parents must allocate more time and energy in telepractice sessions than they do in on-site sessions [33,34].

In this study, the students did not prefer one mode of practice over the other. This could be explained by their satisfaction with telepractice and the significant correlation between their perceptions of telepractice efficacy and their preferences for modes of practice (P<.001). Given that the students had fewer practical concerns than parents, and the fact that students acknowledged the effectiveness of both on-site practice and telepractice, they did not have a preference for the 2 service delivery modes.

Our findings also show that student grade was not significantly associated with telepractice efficacy (parents: P=.76; students: P=.65) or preferences for telepractice and on-site practice (parents: P=.52; students: P=.85). These results reflect the efficacy of telepractice and show that preferences did not differ considerably across different ages. This is consistent with other scientific studies, which have suggested that telepractice is suitable for school-aged students [7-9].

**Study Strengths**

To our knowledge, this study is the first to investigate parents’ and students’ satisfaction with telepractice services for a school-aged population during the COVID-19 pandemic.

Evaluating parents’ and students’ perceptions of the efficacy of telepractice is critical. This information not only helps speech-language therapists understand clients’ perceptions of telepractice, but also informs educational policy makers about the implementation and adoption of telepractice services beyond the pandemic period. Our study clearly demonstrates that users’ satisfaction with telepractice helps to promote evidence-based telepractice. Based on our analysis of both parents’ and students’ attitudes toward telepractice, we believe that both stakeholders acknowledged the efficacy of telepractice. This is a positive indicator for the future adoption of telepractice as another possible service delivery method, which is needed due to the potential psychosocial challenges of the COVID-19 pandemic. Such challenges include disrupted clinical routes, school closures, and reduced educational and medical support [35].

**Limitations and Future Research**

This study focused on a limited sample size with a restricted age range (i.e., Grades 1-7), even though school-based speech therapy services cover students in Grades 1-12. In addition, the small sample size restricted our investigation of the effect of comorbidity on telepractice efficacy, as communication and literacy characteristics can potentially affect telepractice efficacy.

Future research should consider investigating the effect of comorbidity on telepractice efficacy and satisfaction, by testing a larger sample that includes students of different ages and children with different types of special educational needs. For example, parental involvement is lower in the adolescent population than in the younger student population. Furthermore, in the adolescent population, treatment is focused on academic success. It is important to see whether the acceptance of telepractice services among adolescents differs from the acceptance among young, school-aged children. It should also be noted that our study focused on parents’ and students’ satisfaction with telepractice after a relatively short-term telepractice session. Future research should extend this study by investigating parents’ and students’ perceptions of telepractice efficacy and their attitudes toward telepractice after a long-term telepractice session. Our suggestions for future research may elucidate the long-term benefits and sustainability of telepractice, and provide guidance for telepractice strategy development. This information is needed to enhance the quality of digital medical approaches and psychological benefits for children and their families [36].

**Implications**

The results of this study indicate that telepractice efficacy was well acknowledged by parents and students, and that students in Grades 1-7 had similar preferences for telepractice and on-site practice. The use of telepractice is supported not only by scientific evidence, but also by students’ and parents’ satisfaction. These results suggest that telepractice is a possible service delivery option for school-aged students.

The findings of our study are in line with those of existing literature, which suggests that telepractice is a suitable service delivery method [7-9]. Our study provided supporting evidence for schools and speech-language pathologists to adopt...
telepractice in real-life situations. In addition, our results suggest that speech-language pathologists and parents should be more proactive in telepractice services. Given that the parents had a negative view of treatment effectiveness and communication with speech-language pathologists during telepractice sessions, clinicians should consider engaging more effectively with both students and their parents. Speech-language pathologists can regularly update and inform parents and students about treatment effectiveness to increase their confidence during the transition to telepractice. In addition, clinicians should directly address parents' concerns to build a therapeutic relationship [17]. The engagement and participation of parents is highly important in telepractice services. The importance of parent involvement is well noted in the literature [37,38], and the behavioral management of students during telepractice sessions relies on parents. Moreover, the role of the parent in telepractice services extends to providing technical support and troubleshooting [10]. Clinicians can pay attention to potential technical problems and provide relevant support to parents. If clinicians participate in and engage with telepractice services more often, it is expected that parents will have a better rapport with clinicians, which will facilitate the promotion and acceptance of telepractice [37].

Conclusions

This study showed that both Hong Kong Chinese parents and students believed that telepractice was satisfactory and effective. Although students did not prefer one speech therapy delivery mode over the other, parents preferred on-site speech and language therapy. The perceived efficacy of telepractice was associated with students’ preferences for service delivery modes, but it was not associated with parents’ preferences. This could be explained by inadequate communication between clinicians and parents. Our findings suggest that it is necessary for speech-language pathologists to play a more proactive role by integrating telepractice into service delivery and explaining the efficacy of telepractice to parents and students.

Acknowledgments

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Conflicts of Interest

None declared.

Multimedia Appendix 1

Study questionnaire.

[DOCX File , 16 KB - pediatrics_v4i1e25675_app1.docx]

References


Abbreviations

BF: Bayes factor
Interformat Reliability of Web-Based Parent-Rated Questionnaires for Assessing Neurodevelopmental Disorders Among Preschoolers: Cross-sectional Community Study

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Abstract

Background: Early detection and intervention for neurodevelopmental disorders are effective. Several types of paper questionnaires have been developed to assess these conditions in early childhood; however, the psychometric equivalence between the web-based and the paper versions of these questionnaires is unknown.

Objective: This study examined the interformat reliability of the web-based parent-rated version of the Autism Spectrum Screening Questionnaire (ASSQ), Attention-Deficit/Hyperactivity Disorder Rating Scale (ADHD-RS), Developmental Coordination Disorder Questionnaire 2007 (DCDQ), and Strengths and Difficulties Questionnaire (SDQ) among Japanese preschoolers in a community developmental health check-up setting.

Methods: A set of paper-based questionnaires were distributed for voluntary completion to parents of children aged 5 years. The package of the paper format questionnaires included the ASSQ, ADHD-RS, DCDQ, parent-reported SDQ (P-SDQ), and several additional demographic questions. Responses were received from 508 parents of children who agreed to participate in the study. After 3 months, 300 parents, who were among the initial responders, were randomly selected and asked to complete the web-based versions of these questionnaires. A total of 140 parents replied to the web-based format and were included as a final sample in this study.

Results: We obtained the McDonald ω coefficients for both the web-based and paper formats of the ASSQ (web-based: ω=.90; paper: ω=.86), ADHD-RS total and subscales (web-based: ω=.88-.94; paper: ω=.87-.93), DCDQ total and subscales (web-based: ω=.82-.94; paper: ω=.74-.92), and P-SDQ total and subscales (web-based: ω=.55-.81; paper: ω=.52-.80). The intraclass correlation coefficients between the web-based and paper formats were all significant at the 99.9% confidence level: ASSQ (r=0.66, P<.001); ADHD-RS total and subscales (r=0.66-0.74, P<.001); DCDQ total and subscales (r=0.66-0.71, P<.001); P-SDQ Total Difficulties and subscales (r=0.55-0.73, P<.001). There were no significant differences between the web-based and paper formats for total mean score of the ASSQ (P=.76), total (P=.12) and subscale (P=.11-.47) mean scores of DCDQ, and the P-SDQ Total Difficulties mean score (P=.20) and mean subscale scores (P=.28-.79). Although significant differences were found between the web-based and paper formats for mean ADHD-RS scores (total: t132=2.83, P=.005; Inattention subscale: t133=2.15, P=.03; Hyperactivity/Impulsivity subscale: t133=3.21, P=.002), the effect sizes were small (Cohen d=0.18-0.22).

Conclusions: These results suggest that the web-based versions of the ASSQ, ADHD-RS, DCDQ, and P-SDQ were equivalent, with the same level of internal consistency and intrarater reliability as the paper versions, indicating the applicability of the web-based versions of these questionnaires for assessing neurodevelopmental disorders.
neurodevelopmental disorders; web-based questionnaire; preschoolers; parents; interformat reliability

Introduction

In the American Psychiatric Association's Diagnostic and Statistical Manual for Mental Disorders, Fifth Edition (DSM-5), neurodevelopmental disorders are identified in the early developmental stages and are characterized by developmental deficits that lead to impairments in personal, social, academic, and vocational functioning [1]. Representative examples of such disorders include autism spectrum disorder, attention-deficit/hyperactivity disorder, and developmental coordination disorder. The core characteristics of autism spectrum disorder include 2 main dimensions—social communication and restricted, repetitive sensory–motor behaviors—that are irrespective of culture, race, ethnicity, or socioeconomic group [2]. Estimates of the total-population prevalence of autism spectrum disorder range from 2.2% to 3.2% [3,4]. The hallmarks of attention-deficit/hyperactivity disorder are developmentally impaired attention, motor hyperactivity, impulsivity, and the difficulties associated with them [5]. A recent meta-analysis [6] revealed that the estimated prevalence of children with attention-deficit/hyperactivity disorder is 3.4% in the general population. Developmental coordination disorder is characterized by marked impairment in the acquisition and execution of motor skills. This impairment significantly and sustainably interferes with activities of daily living, including academic achievement [1]. A recent review [7] reported that prevalence estimates for developmental coordination disorder among children range from 2% to 20%, with 5% to 6% being the most commonly reported prevalence rate.

It is known that children with these conditions not only have various secondary mental health problems [8-10] but also experience maladjustment in adulthood [11-13]. Since a number of studies [7,14,15] have reported that early detection and intervention for neurodevelopmental disorders is effective, it is necessary to develop useful screening tools for assessing these conditions in early childhood.

Several questionnaires have been developed to assess a variety of neurodevelopmental disorders. The Autism Spectrum Screening Questionnaire (ASSQ) was developed to screen for autism spectrum disorder in school-age children based on their parents’ or primary caregivers’ ratings [16] and has been shown to be highly accurate in screening for autism spectrum disorder [17]. Additional research has confirmed that the ASSQ has good reliability and validity and can be applied to preschool-age children, as well [18,19]. The ADHD-Rating Scale (ADHD-RS) is one of the most widely used questionnaires developed to assess ADHD symptoms in children age 5 to 18 years [20]. There are 2 versions of the ADHD-RS: the home form is rated by parents or primary caregivers of the children, and the school form is rated by teachers. It has been demonstrated that the ADHD-RS has good reliability and validity in preschool children [21]. Moreover, previous research has confirmed that the ADHD-RS shows higher sensitivity and specificity in parent ratings than those in teacher ratings among preschoolers [22]. The Developmental Coordination Disorder Questionnaire 2007 (DCDQ) was developed to identify children age 5 to 15 years who are at risk for developmental coordination disorder, based on parents’ ratings [23]. It has been found that the DCDQ has good psychometric properties and has been recommended for use in clinical practice as supplemental information for the diagnosis of children with developmental coordination disorder [7]. It is known that children with neurodevelopmental disorders have behavioral and emotional difficulties. The Strengths and Difficulties Questionnaire (SDQ) is a brief behavioral screening questionnaire about externalizing and internalizing problems in children [24]. It has been reported that using parent ratings for the SDQ has satisfactory reliability and validity in a community sample of 5- to 15-year-old children [25].

These questionnaires have been developed as paper-and-pencil type questionnaires and can be useful in individual clinical settings; however, to screen large populations in a local community for early detection of neurodevelopmental disorders, web-based versions are more efficient than the paper-and-pencil version because of the significant time and effort required to distribute and collect paper-and-pencil questionnaires. Not only can costs be saved, but also there are additional advantages; for example, we can automate the process of manual data entry after completing data collection [26]. In addition, using web-based questionnaires to collect data generally improves the quality of the data because the validation checks can incorporate prompts that alert respondents if they enter incorrect or incomplete answers [27]. Furthermore, web-based instruments expand the reach of assessments, which is particularly important under pandemic conditions when it is difficult or impossible to administer in-person assessments [28,29].

On one hand, several studies [30-32] have reported psychometric equivalence between web-based and paper-and-pencil versions of the questionnaires used to assess various psychological disorders; on the other hand, some studies have revealed psychometrically significant differences between the 2 formats [26,33,34]. Therefore, it is necessary to evaluate the comparability of web-based and paper-and-pencil versions of the questionnaires used to assess various psychological disorders; on the other hand, some studies have revealed psychometrically significant differences between the 2 formats [26,33,34]. Therefore, it is necessary to evaluate the comparability of web-based and paper-and-pencil versions of the questionnaires [35]. In particular, high interformat reliability, meaning the level of equality between different delivery formats, indicates that the psychometric properties of the instrument are independent of the delivery format [36]. However, to our knowledge, no studies have assessed the interformat reliability of web-based questionnaires that aim to assess neurodevelopmental disorders.

We aimed to examine the interformat reliability of the web-based versions of the ASSQ, ADHD-RS, DCDQ, and SDQ. Based on previous work [36], we confirmed interformat reliability from the following 3 perspectives. First, we verified the internal consistency of the web-based and paper-and-pencil formats of each questionnaire. Second, we examined the
intraclass correlations between the 2 formats of each questionnaire to test intrarater reliability. Third, we investigated the mean score differences between the 2 formats of each questionnaire to confirm equivalence in quality.

**Methods**

**Participants**

This study was conducted as part of the Hirosaki Five-Year-Old Children Developmental Health Check-up Study (HFC Study), a large community-based cohort study initiated in 2013 that examined the impact of children’s neurodevelopmental disorders and lifestyle habits on their adaptation and emotional and behavioral problems at age 5 years. Located in Aomori Prefecture in the northeastern part of Japan, Hirosaki City has approximately 175,000 residents, 1 university, and several colleges, and its main industry is agriculture.

Participants in this study were recruited in July 2018. The local government of Hirosaki City distributed a set of paper-based questionnaires for voluntary completion to the parents of 620 5-year-old children in the city via the municipal health center. The package included the ASSQ, ADHD-RS, DCDQ, parent-reported SDQ (P-SDQ), and demographic questions. Responses were received from 508 parents who agreed to participate in the study. After 3 months, 300 of the 508 respondents were randomly selected and informed of the objective of this study. The individuals who gave their written consent to participate were asked to complete web-based versions of these questionnaires. Participants were given an ID and password to complete the web-based survey on their own computers. There were no restrictions on the type of computer (eg, personal computer, tablet, or smartphone) that they could use to complete the survey. A total of 140 parents replied to the web-based format and were included in the final sample in the present study. Table 1 shows the demographic characteristics of this sample; Multimedia Appendix 1 contains the characteristics of the 368 people who did not respond to the web-based survey.

**Table 1.** Participants’ demographic characteristics.

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>Value (N=140), n (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Children’s gender</td>
<td></td>
</tr>
<tr>
<td>Boy</td>
<td>78 (55.7)</td>
</tr>
<tr>
<td>Girl</td>
<td>62 (44.3)</td>
</tr>
<tr>
<td>Children’s age (months)</td>
<td></td>
</tr>
<tr>
<td>60</td>
<td>3 (2.1)</td>
</tr>
<tr>
<td>61</td>
<td>25 (17.9)</td>
</tr>
<tr>
<td>62</td>
<td>21 (15.0)</td>
</tr>
<tr>
<td>63</td>
<td>25 (17.9)</td>
</tr>
<tr>
<td>64</td>
<td>19 (13.6)</td>
</tr>
<tr>
<td>65</td>
<td>32 (22.9)</td>
</tr>
<tr>
<td>66</td>
<td>15 (10.7)</td>
</tr>
<tr>
<td>Respondent</td>
<td></td>
</tr>
<tr>
<td>Mother</td>
<td>127 (90.7)</td>
</tr>
<tr>
<td>Father</td>
<td>13 (9.3)</td>
</tr>
<tr>
<td>Childcare during daytime</td>
<td></td>
</tr>
<tr>
<td>Nursery school</td>
<td>115 (82.1)</td>
</tr>
<tr>
<td>Kindergarten</td>
<td>24 (17.1)</td>
</tr>
<tr>
<td>Mother</td>
<td>1 (0.7)</td>
</tr>
<tr>
<td>Household income (JPY(^a))</td>
<td></td>
</tr>
<tr>
<td>&lt;2 million</td>
<td>10 (7.1)</td>
</tr>
<tr>
<td>2-4 million</td>
<td>44 (31.4)</td>
</tr>
<tr>
<td>4-7 million</td>
<td>56 (40.0)</td>
</tr>
<tr>
<td>7-10 million</td>
<td>18 (12.9)</td>
</tr>
<tr>
<td>&gt;10 million</td>
<td>7 (5.0)</td>
</tr>
<tr>
<td>Don’t know</td>
<td>5 (3.6)</td>
</tr>
</tbody>
</table>

\(^a\)JPY: Japanese Yen; an approximate exchange rate of US $1= 103.80 JPY.
Measures

ASSQ

The ASSQ has 27 items that assess autistic features such as social interaction and communication problems, behaviors that are restrictive and repetitive, motor clumsiness, and other associated symptoms, including motor and vocal tics [16,17]. The items are rated on a 3-point scale ranging from 0 (not true) to 2 (true). A higher ASSQ score indicates more severe autistic problems. The total possible score of the ASSQ ranges from 0 to 54. In this study, we used the Japanese version of the ASSQ. A previous study [19] revealed that the ASSQ had good reliability (autism spectrum disorder clinical group: Cronbach $\alpha$=.88; community group: Cronbach $\alpha$=.87) and validity as a screening instrument for use with preschoolers in Japanese community settings.

ADHD-RS

The ADHD-RS includes 18 items to measure 2 features of attention-deficit/hyperactivity disorder: Inattention (9-item subscale) and Hyperactivity/Impulsivity (9-item subscale) [20]. It is evaluated on a 4-point scale ranging from 0 (not at all or rarely) to 3 (very often). Higher scores on the ADHD-RS indicate more severe attention-deficit/hyperactivity disorder problems, with total scores ranging from 0 to 54. This study used the Japanese version of the ADHD-RS home form [37]. A previous study [22] revealed that this version of the ADHD-RS had sufficient reliability (Inattention subscale: Cronbach $\alpha$=.88, Hyperactivity/Impulsivity subscale: Cronbach $\alpha$=.85) and validity for screening children potentially living with attention-deficit/hyperactivity disorder in a community setting.

DCDQ

The DCDQ consists of 15 items organized into 3 subscales—Control During Movements (6 items), Fine Motor and Handwriting (4 items), and General Coordination (5 items) [23]. Parents or primary caregivers were asked to evaluate the degree of motor coordination in their children compared to that of other children of the same age on a 5-point scale ranging from 1 (not at all like your child) to 5 (extremely like your child). Lower scores indicate severe developmental coordination disorder symptoms. The total possible score ranges from 15 to 75. This study used the Japanese version of the DCDQ, which has sufficient criterion validity, fit indices, and internal consistency (Total: Cronbach $\alpha$=.93; Control During Movements subscale: Cronbach $\alpha$=.91; Fine Motor and Handwriting: Cronbach $\alpha$=.91; General Coordination: Cronbach $\alpha$=.81) when used with preschool- and school-age children [38].

P-SDQ

The P-SDQ includes 25 items that assess children’s strengths and difficulties on 5 different subscales (each comprising 5 items): Emotional Symptoms, Conduct Problems, Hyperactivity/Inattention, Peer Relationship Problems, and Prosocial Behavior [24,25]. Parents or principal caregivers rated the items on a 3-point scale ranging from 0 (not true) to 2 (certainly true). The score for each subscale is calculated by summing the scores of 5 items, ranging from 0 to 10. The Total Difficulties score is calculated by summing the 4 difficulty subscale scores, ranging from 0 to 40. Higher scores on the 4 difficulty subscales as well as the Total Difficulties score indicate more severe emotional and behavioral deficits. Meanwhile, a higher score on the Prosocial Behavior subscale represents a more positive aspect of prosocial behavior. In this study, we used the P-SDQ, which showed favorable psychometric properties in Japanese community-based samples (Total Difficulties: Cronbach $\alpha$ =.77; Emotional Symptoms: Cronbach $\alpha$ =.61; Conduct Problems: Cronbach $\alpha$ =.52; Hyperactivity/Inattention: Cronbach $\alpha$ =.75; Peer Relationship Problems: Cronbach $\alpha$ =.52; Prosocial Behavior: Cronbach $\alpha$ =.69) [39].

Statistical Analysis

To test internal consistency, we calculated McDonald $\omega$ coefficients for the total and subscale scores of each measure, based on a previous study’s recommendation [40], for both web-based and paper formats. We also calculated intraclac correlation coefficients between the web-based and paper formats to evaluate the interformat reliability. Paired 2-tailed $t$ tests were performed to evaluate mean score differences between the web-based and paper formats to examine equivalence in quality. A $P$ value <.05 was statistically significant. Analyses were performed using SPSS software (version 25.0; IBM Corp) and R (version 4.0.3; R Foundation for Statistical Computing).

Ethics

The research was performed in accordance with the ethical guidelines of the Declaration of Helsinki. The protocol of this study was approved by the Committee on Medical Ethics of Hirosaki University (IRB 2018-168). To protect personal data, we adhered to the city’s and the committee’s information security policies.

Results

Internal Consistency

Table 2 shows the McDonald $\omega$ coefficients for both formats of the questionnaires.
Table 2. McDonald $\omega$ coefficients for the web-based and paper versions of the ASSQ, ADHD-RS, DCDQ, and P-SDQ (N=140).

<table>
<thead>
<tr>
<th>Scale and subscales</th>
<th>McDonald $\omega$</th>
<th>Web-based</th>
<th>Paper</th>
</tr>
</thead>
<tbody>
<tr>
<td>Autism Spectrum Screening Questionnaire</td>
<td></td>
<td>.90</td>
<td>.86$^a$</td>
</tr>
<tr>
<td>Attention-Deficit/Hyperactivity Disorder Rating Scale</td>
<td></td>
<td>.94</td>
<td>.93$^b$</td>
</tr>
<tr>
<td>Total</td>
<td></td>
<td>.90</td>
<td>.88$^c$</td>
</tr>
<tr>
<td>Inattention</td>
<td></td>
<td>.88</td>
<td>.87$^c$</td>
</tr>
<tr>
<td>Hyperactivity/Impulsivity</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Developmental Coordination Disorder Questionnaire</td>
<td></td>
<td>.94</td>
<td>.92$^a$</td>
</tr>
<tr>
<td>Total</td>
<td></td>
<td>.87</td>
<td>.86$^a$</td>
</tr>
<tr>
<td>Control During Movement</td>
<td></td>
<td>.88</td>
<td>.91$^d$</td>
</tr>
<tr>
<td>Fine Motor/Handwriting</td>
<td></td>
<td>.82</td>
<td>.74</td>
</tr>
<tr>
<td>General Coordination</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Parent-rated Strength and Difficulties Questionnaire</td>
<td></td>
<td>.81</td>
<td>.78</td>
</tr>
<tr>
<td>Total Difficulties</td>
<td></td>
<td>.64</td>
<td>.70</td>
</tr>
<tr>
<td>Emotional Symptoms</td>
<td></td>
<td>.55</td>
<td>.50</td>
</tr>
<tr>
<td>Conduct Problems</td>
<td></td>
<td>.78</td>
<td>.79</td>
</tr>
<tr>
<td>Hyperactivity/Inattention</td>
<td></td>
<td>.57</td>
<td>.52</td>
</tr>
<tr>
<td>Peer Relationship Problems</td>
<td></td>
<td>.76</td>
<td>.80</td>
</tr>
<tr>
<td>Prosocial Behavior</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

$^a$Calculated for 137 participants because of missing data.
$^b$Calculated for 133 participants because of missing data.
$^c$Calculated for 134 participants because of missing data.
$^d$Calculated for 139 participants because of missing data.

Based on a previous study [41], an internal consistency coefficient below .70 is considered unacceptable, a coefficient from .70 to .79 is considered fair, a coefficient from .80-.89 is considered good, and a coefficient of .90 or above is considered excellent. The McDonald $\omega$ coefficients for both the web-based and paper formats of the ASSQ ranged from .86 to .90, indicating good to excellent internal consistency. The McDonald $\omega$ coefficients for both the web-based and paper formats of the overall ADHD-RS and its subscales ranged from .87 to .94, indicating fair to excellent internal consistency. The McDonald $\omega$ coefficients for the web-based and paper formats of the Total Difficulties subscale and the subscales of the P-SDQ ranged from .52 to .81. Notably, the McDonald $\omega$ coefficients for both the web-based and paper versions of the Peer Relationship Problems and Conduct Problems subscales and the web-based version of the Emotional Symptoms subscale were all unacceptable [41], with coefficients ranging from .51 to .66.

Intraclass Correlation Coefficients

Table 3 presents the intraclass correlation coefficients between each format for ASSQ, ADHD-RS, DCDQ, and P-SDQ.
Table 3. Intraclass correlation coefficients between the web-based and paper formats of the ASSQ, ADHD-RS, DCDQ, and P-SDQ (N=140).

<table>
<thead>
<tr>
<th>Scale and subscales</th>
<th>Intraclass correlation&lt;sup&gt;a&lt;/sup&gt; (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Autism Spectrum Screening Questionnaire</td>
<td>0.66&lt;sup&gt;b&lt;/sup&gt; (0.56-0.75)</td>
</tr>
<tr>
<td><strong>Attention-Deficit/Hyperactivity Disorder Rating Scale</strong></td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>0.72&lt;sup&gt;c&lt;/sup&gt; (0.62-0.79)</td>
</tr>
<tr>
<td>Inattention</td>
<td>0.66&lt;sup&gt;d&lt;/sup&gt; (0.55-0.74)</td>
</tr>
<tr>
<td>Hyperactivity/Impulsivity</td>
<td>0.74&lt;sup&gt;d&lt;/sup&gt; (0.65-0.80)</td>
</tr>
<tr>
<td><strong>Developmental Coordination Disorder Questionnaire</strong></td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>0.71&lt;sup&gt;b&lt;/sup&gt; (0.61-0.78)</td>
</tr>
<tr>
<td>Control During Movement</td>
<td>0.71&lt;sup&gt;b&lt;/sup&gt; (0.62-0.79)</td>
</tr>
<tr>
<td>Fine Motor/Handwriting</td>
<td>0.66&lt;sup&gt;d&lt;/sup&gt; (0.55-0.74)</td>
</tr>
<tr>
<td>General Coordination</td>
<td>0.66 (0.56-0.75)</td>
</tr>
<tr>
<td><strong>Parent-rated Strength and Difficulties Questionnaire</strong></td>
<td></td>
</tr>
<tr>
<td>Total Difficulties</td>
<td>0.73 (0.65-0.80)</td>
</tr>
<tr>
<td>Emotional Symptoms</td>
<td>0.59 (0.47-0.69)</td>
</tr>
<tr>
<td>Conduct Problems</td>
<td>0.66 (0.55-0.74)</td>
</tr>
<tr>
<td>Hyperactivity/Inattention</td>
<td>0.68 (0.58-0.76)</td>
</tr>
<tr>
<td>Peer Relationship Problems</td>
<td>0.58 (0.45-0.68)</td>
</tr>
<tr>
<td>Prosocial Behavior</td>
<td>0.55 (0.43-0.66)</td>
</tr>
</tbody>
</table>

<sup>a</sup>All correlations were significant at the P<.001 level.

<sup>b</sup>Calculated for 137 participants because of missing data.

<sup>c</sup>Calculated for 133 participants because of missing data.

<sup>d</sup>Calculated for 134 participants because of missing data.

<sup>e</sup>Calculated for 139 participants because of missing data.

Intraclass correlation coefficients between 0.50 and 0.75 are considered moderate, whereas values above 0.75 are considered high [42]. The intraclass correlation coefficient between the web-based and paper formats of ASSQ was moderate and significant (P<.001). There were also moderate significant (P<.001) intraclass correlations found between the web-based and paper formats of the overall scale and subscales of the ADHD-RS and the DCDQ, and the P-SDQ subscales.

### Mean Differences Between the Web-Based and Paper-and-Pencil Formats

Table 4 shows the mean scores and standard deviations of both formats of the ASSQ, ADHD-RS, DCDQ, and P-SDQ.
The purpose of this study was to examine the interformat reliability of the web-based versions of the ASSQ, ADHD-RS, DCDQ, and SDQ by comparing the internal consistency, intraclass correlation, and mean score differences of their web-based and paper formats.

For the ASSQ, ADHD-RS, and DCDQ, the McDonald $\omega$ coefficients were sufficient for both the web-based and paper versions, similar to findings in previous studies [19,22,38] that calculated Cronbach $\alpha$. These results indicate that the web-based format of these questionnaires has good internal consistency. The McDonald $\omega$ coefficients for the Total Difficulties, Hyperactivity/Inattention, and Prosocial Behavior subscales of both the web-based and paper versions of the P-SDQ were also good. The McDonald $\omega$ coefficient for the paper version of the Emotional Symptoms subscale in this study was also good; however, it was unsatisfactory for the Conduct Problems and Peer Relationship Problems subscales of both the web-based and paper versions. The McDonald $\omega$ coefficient for the web-based version of the Emotional Symptoms subscale was also relatively low. These results are similar to those found in studies [25,39,43] that calculated Cronbach $\alpha$ for the paper format versions. Therefore, our findings suggest that, similar to the paper format, there are a few difficulties in using the web-based format of the P-SDQ for assessing externalizing and internalizing problems in children.

We found that there were significant ($P<.001$) moderate positive intraclass correlations between the web-based and paper formats of the ASSQ, ADHD-RS, DCDQ total scores, and Total Difficulties score of the P-SDQ. Similar to this study, several earlier studies [44-46] on the equivalence between web-based and paper formats of self-report questionnaires meant to assess psychiatric symptoms have reported moderate significant correlations, suggesting that web-based questionnaire administration was a reliable alternative to using the paper format. Hence, it seems that the questionnaires assessing neurodevelopmental disorders, such as the ASSQ, ADHD-RS, DCDQ, and P-SDQ, can be made available and administered in web-based situations as well. However, another study [27] had earlier pointed out that the agreement rate between the web-based and paper formats was higher for objective factual questions than for questions based on personal subjective evaluation. The questionnaires used in this study were subjective evaluations of children’s developmental status by parents, which may have impacted the correlation values.

**Discussion**

**Principal Findings**

The purpose of this study was to examine the interformat reliability of the web-based versions of the ASSQ, ADHD-RS, DCDQ, and SDQ by comparing the internal consistency, intraclass correlation, and mean score differences of their web-based and paper formats. Table 4. Mean scores for the web-based and paper formats of ASSQ, ADHD-RS, DCDQ, and P-SDQ (N=140).

<table>
<thead>
<tr>
<th>Scale and subscales</th>
<th>Web-based, mean (SD)</th>
<th>Paper, mean (SD)</th>
<th>$t$ test (df)</th>
<th>$P$ value</th>
<th>Cohen $d$</th>
</tr>
</thead>
<tbody>
<tr>
<td>Autism Spectrum Screening Questionnaire</td>
<td>4.10 (5.46)</td>
<td>3.99 (4.67)</td>
<td>0.31 (136)</td>
<td>.76</td>
<td>0.02</td>
</tr>
<tr>
<td><strong>Attention-Deficit/Hyperactivity Disorder Rating Scale</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>4.88 (7.08)</td>
<td>6.14 (6.94)</td>
<td>2.83 (132)</td>
<td>.005</td>
<td>0.18</td>
</tr>
<tr>
<td>Inattention</td>
<td>2.64 (3.84)</td>
<td>3.22 (3.69)</td>
<td>2.15 (133)</td>
<td>.03</td>
<td>0.22</td>
</tr>
<tr>
<td>Hyperactivity/Impulsivity</td>
<td>2.22 (3.46)</td>
<td>2.92 (3.62)</td>
<td>3.22 (136)</td>
<td>.002</td>
<td>0.20</td>
</tr>
<tr>
<td><strong>Developmental Coordination Disorder Questionnaire</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total</td>
<td>58.85 (11.53)</td>
<td>57.74 (10.52)</td>
<td>1.55 (136)</td>
<td>.12</td>
<td>0.09</td>
</tr>
<tr>
<td>Control During Movement</td>
<td>22.69 (4.89)</td>
<td>22.21 (4.48)</td>
<td>1.57 (136)</td>
<td>.12</td>
<td>0.06</td>
</tr>
<tr>
<td>Fine Motor/Handwriting</td>
<td>16.54 (3.41)</td>
<td>16.36 (3.66)</td>
<td>0.73 (138)</td>
<td>.47</td>
<td>0.05</td>
</tr>
<tr>
<td>General Coordination</td>
<td>19.72 (4.35)</td>
<td>19.26 (3.95)</td>
<td>1.61 (139)</td>
<td>.11</td>
<td>0.11</td>
</tr>
<tr>
<td><strong>Parent-rated Strength and Difficulties Questionnaire</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total Difficulties</td>
<td>7.42 (4.83)</td>
<td>7.80 (4.72)</td>
<td>1.29 (139)</td>
<td>.20</td>
<td>0.08</td>
</tr>
<tr>
<td>Emotional Symptoms</td>
<td>1.74 (1.69)</td>
<td>1.88 (1.76)</td>
<td>1.08 (139)</td>
<td>.28</td>
<td>0.07</td>
</tr>
<tr>
<td>Conduct Problems</td>
<td>1.82 (1.50)</td>
<td>1.94 (1.51)</td>
<td>1.08 (139)</td>
<td>.28</td>
<td>0.08</td>
</tr>
<tr>
<td>Hyperactivity/Inattention</td>
<td>2.67 (2.16)</td>
<td>2.76 (2.17)</td>
<td>0.63 (139)</td>
<td>.53</td>
<td>0.05</td>
</tr>
<tr>
<td>Peer Relationship Problems</td>
<td>1.19 (1.37)</td>
<td>1.22 (1.36)</td>
<td>0.27 (139)</td>
<td>.79</td>
<td>0.02</td>
</tr>
<tr>
<td>Prosocial Behavior</td>
<td>7.86 (1.92)</td>
<td>7.77 (2.12)</td>
<td>0.53 (139)</td>
<td>.60</td>
<td>0.05</td>
</tr>
</tbody>
</table>

There was no significant difference between the web-based and paper formats for the total mean score of the ASSQ ($P=.76$). Web-based scores were significantly lower than those of the paper format for the total mean scores of the ADHD-RS ($t_{132}=2.83$, $P=.005$, Cohen $d=0.18$) and for its subscales (Inattention: $t_{133}=2.15$, $P=.03$, Cohen $d=0.22$; Hyperactivity/Impulsivity: $t_{133}=3.21$, $P=.002$, Cohen $d=0.20$). We found no significant differences between the web-based and paper formats for the mean scores on the DCDQ total ($P=.12$) and subscales (Control during Movement: $P=.12$; Fine Motor/Handwriting: $P=.47$; General Coordination: $P=.11$). Similarly, there were no significant differences between the web-based and paper formats for total P-SDQ scores ($P=.20$) and mean subscale scores (Emotional Symptoms: $P=.28$; Conduct Problems: $P=.28$; Hyperactivity/Inattention: $P=.53$; Peer Relationship Problems: $P=.79$; Prosocial Behavior: $P=.60$).
Furthermore, the analyses revealed that there were no significant differences in the ASSQ total mean scores between the web-based and paper formats (P=0.76). This result suggests that the web-based version of the ASSQ is equal in quality to that of the paper version. However, there were significant differences in the ADHD-RS total and subscale mean scores between the web-based and paper formats (total: P=0.005; Inattention: P=0.03; Hyperactivity/Impulsivity: P=0.002). Previous studies [33,36,47] have reported a significant difference in mean scores between web-based and paper formats; however, due to the small effect size (Cohen d=0.14-0.27), it was determined that the statistically significant difference in mean scores was not clinically meaningful in practice. In light of this finding, web-based questionnaires are a potential substitute for paper-based questionnaires. The effect sizes obtained in this study were also small (Cohen d=0.18-0.22). Furthermore, the web-based version McDonald to values in this study were slightly higher than those of the paper format. These results suggest that the ADHD-RS is applicable for web-based utilization. We found that there were no significant differences in the DCDQ total and subscale mean scores between the web-based and paper formats (total: P=0.12; Control during Movement: P=0.12; Fine Motor/Handwriting: P=0.47; General Coordination: P=0.11). Additionally, we also confirmed that there were no significant differences in the P-SDQ total and subscale mean scores between the web-based and paper formats (total: P=0.20; Emotional Symptoms: P=0.28; Conduct Problems: P=0.28; Hyperactivity/Inattention: P=0.53; Peer Relationship Problems: P=0.79; Prosocial Behavior: P=0.60). These results suggest that both the DCDQ and P-SDQ web-based formats are equivalent in quality to those of the paper format.

**Strengths and Limitations**

The evidence found in this study supports the applicability of the web-based versions of the ASSQ, ADHD-RS, DCDQ, and P-SDQ. It has been pointed out that children with possible neurodevelopmental disorders may be overlooked during developmental health check-ups in Japan [48], and there is a lack of specialized organizations capable of assessing neurodevelopmental disorders. Furthermore, in the current COVID-19 pandemic, it is also difficult to conduct in-person evaluations. This study, which shows the applicability of web-based questionnaires assessing neurodevelopmental disorders, has the potential to improve early detection and intervention for these disorders in regions where specialized services are lacking and under the present pandemic conditions.

However, there are some limitations to this research. First, the discriminant validity of the web-based version of each questionnaire was not confirmed in this study. This is because this study used a cross-sectional research design as part of a community developmental health check-up. Therefore, it is unclear whether the children included in this study had been diagnosed with neurodevelopmental disorders or experienced other emotional or behavioral problems or deficits that do not meet the criteria for a clinical diagnosis. It is necessary to verify the discriminant ability of the web-based version of each questionnaire for both clinical and nonclinical groups. Second, the raters who evaluated the children’s condition in this study were mostly parents. Previous research [17,49,50] conducted with paper-based questionnaires has examined the psychometric properties of the teacher-rated version of each questionnaire that was used in this study. In the future, we also need to clarify the psychometric properties of the teacher-rated web-based version of the questionnaires used in this study compared to the parent-rated web-based version. Third, we were not able to examine the impact of the order in which the questionnaires were administered. Previous studies [26,33] have confirmed that the order in which web- and paper-based questionnaires are administered has an effect on the scores of those questionnaires. In the future, it will be necessary to investigate the effect of the order of administration of the questionnaires in the research design. Fourth, the age of the children in this study was limited to 5 years. Previous studies [17,38,49,50] on the paper-based ASSQ, ADHD-RS, and SDQ have been conducted with school-age children. It is necessary to investigate the psychometric properties of the web-based version of the ASSQ, ADHD-RS, DCDQ, and P-SDQ among school-age children. Fifth, we did not control for the computer used by the participants in this study. We need to test whether the type of computer affects their responses in the future. Finally, this study was conducted in one medium-size city in Japan, thereby limiting the generalizability of its findings to other regions.

**Conclusions**

This study examined the interformat reliability of the web-based versions of questionnaires for assessing neurodevelopmental disorders. Our findings showed that the web-based versions of the ASSQ, ADHD-RS, DCDQ, and P-SDQ had the same level of internal consistency, intrarater reliability, and equality as their paper versions. These results indicate the web applicability of these questionnaires for assessing neurodevelopmental disorders.

**Acknowledgments**

M Tanaka and MS conceptualized and designed the study. Data collection was conducted by M Tanaka and MS. M Tanaka conducted data analysis and drafted the initial manuscript. MS, M Takahashi, and MA reviewed the manuscript and approved the final manuscript as submitted. KN conceptualized and designed the study, critically reviewed the manuscript, and approved the final manuscript as submitted. MS, M Takahashi, MA, and KN administered developmental check-ups for children who participated in the study. All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work. This study was conducted by the Graduate School of Medicine at Hirosaki University, in close collaboration with the municipal health center and the city. We express gratitude to all the participants and their families. The authors gratefully acknowledge the contributions of local practitioners, public servants, and students. This study was financially supported by the

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Conflicts of Interest
None declared.

Multimedia Appendix 1
Participants who took part in the paper-and-pencil survey only (N=368).

References


40. McNeish D. Thanks coefficient alpha, we’ll take it from here. Psychol Methods 2018 Sep;23(3):412-433. [doi: 10.1037/met0000144] [Medline: 28557467]


**Abbreviations**

ADHD-RS: Attention-Deficit/Hyperactivity Disorder Rating Scale  
ASSQ: Autism Spectrum Screening Questionnaire  
DCDQ: Developmental Coordination Disorder Questionnaire 2007  
DSM-5: Diagnostic and Statistical Manual for Mental Disorders (Fifth Edition)  
P-SDQ: Parent-reported Strengths and Difficulties Questionnaire  
SDQ: Strengths and Difficulties Questionnaire

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Monitoring Adherence Rate to Growth Hormone Therapy and Growth Outcomes in Taiwanese Children Using Easypod Connect: Observational Study

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Abstract

Background: Adherence to growth hormone therapy is difficult to detect reliably. Devices such as easypod have been developed for electronic recording of injections. The easypod connect observational study (ECOS) was an open-label, observational, multinational, phase IV study conducted in 24 countries around the world. The final results from ECOS in the Taiwanese cohort are reported in this paper.

Objective: This study aimed to evaluate the adherence and long-term outcomes of growth hormone therapy in pediatric subjects using the easypod electromechanical device.

Methods: Subjects (aged 2-18 years or >18 years without fusion of growth plates) who received Saizen (recombinant human growth hormone, somatropin) via the easypod device were enrolled in this study. The primary objective was to assess the level of adherence in subjects receiving Saizen via easypod.

Results: In Taiwan, a total of 35 and 13 children fulfilled the criteria of full analysis set and complete analysis set, respectively. The mean (SD) age of the complete analysis set was 12.08 (2.72) years. All subjects were growth hormone–naïve, with 38% (5/13) females. The mean adherence rates of 13 subjects were 87.6% at 3 months and 84.3% at 6 months, that of 8 subjects was 81.0% at 9 months, and that of 4 subjects was 91.6% at 1 year. After 1 year of treatment, subjects had a median (Q1:Q3) change in height SD score of 0.30 (0.06:0.48), median height velocity of 6.50 (4.33:8.24) cm/year, and median change in height velocity SD score of 1.81 (–0.04:3.52).

Conclusions: With the easypod device, patients with inadequate adherence and poor response to treatment can be identified. Adherence to growth hormone therapy administered via easypod was generally high in the first year of treatment but the adherence gradually decreased over time. Overall, growth outcomes after 1 year indicated a positive growth response to growth hormone treatment. Future efforts should be focused on personalized management of adherence by using the easypod system.


KEYWORDS

growth hormone; adherence; easypod; eHealth
Introduction

Background

Human growth hormone, also known as somatotropin, is synthesized and secreted by the somatotropic cells of the anterior pituitary gland and it plays a critical role in growth and metabolism. Recombinant human growth hormone was first approved for the treatment of childhood growth hormone deficiency in 1985 [1]. Since then, synthetic human growth hormone has been widely administered for the treatment of inadequate secretion of endogenous growth hormones in children and adults. For pediatric patients, growth hormone is indicated for treating growth disorders due to a number of medical causes, including growth hormone deficiency, Turner syndrome, and children born small for gestational age. During the past several decades, growth hormone therapy has demonstrated its effects on improving growth outcomes and helping children achieve catch-up growth [2-5].

Adherence to Growth Hormone Treatment

As growth hormone therapy for children generally starts at a young age and lasts for several years, both the child and the family are involved in this long-term treatment process. For chronic non–life-threatening conditions such as growth hormone deficiency [6], adherence to treatment is relatively difficult to maintain at a high level, especially when the benefits are not immediately apparent, and regular subcutaneous injections with a frequency of up to once daily causes both physical and psychological burdens. Even though adherence can be monitored through methods such as diary cards or by comparing total expected growth hormone usage to the total amount of growth hormone prescribed, the data could easily be overestimated and become unreliable since the child or the parents may be reluctant to admit missing injections [7].

Studies have shown that growth outcomes of growth hormone therapy could be affected by multiple factors [8-10], among which, poor adherence is still a major problem in treating growth disorders for pediatric patients [11,12]. Although the results vary substantially between studies due to the methods and definitions applied, a prevalence of 5%–82% has been reported [13]. Poor adherence not only results in suboptimal growth but also increases unnecessary medical expenses [14,15].

The frequency of inadequate adherence is usually underestimated when assessed using conventional methods (eg, diary cards, questionnaires, number of returned vials) [13,16], which only give fragmentary pictures of a patient’s dosing history. In addition, the aforementioned methods cannot completely reveal the patterns of nonadherence such as reduced dosage, drug holiday, or delayed initiation [16]. Thus, electronic monitoring of drug dosing histories is currently recognized as a standard for adherence quantification [16]. The electronic monitoring of injections via devices such as easypod provides information on how many doses have been taken as prescribed and about nonadministered doses, thereby reflecting the extent to which the patient is adherent to the therapy. Through adequate monitoring methods, physicians are able to promptly evaluate the adherence following an inadequate response to growth hormone therapy [17,18].

Objectives

The easypod connect observational study (ECOS) was an open-label, observational, longitudinal study conducted in 24 countries (Argentina, Australia, Austria, Canada, China, Colombia, Czech Republic, Finland, France, Greece, Hungary, Indonesia, Italy, Kingdom of Saudi Arabia, Korea, Mexico, Norway, Singapore, Slovakia, Spain, Sweden, Taiwan, United Arab Emirates, and the United Kingdom) with a total of 1203 subjects included for analyses. It aimed to evaluate the adherence and long-term outcomes of therapy in pediatric subjects using the easypod electromechanical device for growth hormone treatment and to undertake population-based analyses to generate hypotheses relating to drivers of individual adherence [19]. The results of the ECOS have been published by Koledova et al [19]. Among the countries involved, the results of Spain [20], Italy [21], and Mexico [22] have been published. However, there is no related publication in the Asia-Pacific region. The culture and living habits might possibly influence medication adherence. Clinically, some children in Taiwan go to bed late, resulting in late administration of the growth hormone, which may indirectly affect adherence. Therefore, in this study, we present the results of Taiwanese pediatric subjects.

Methods

Study Design

ECOS was a multinational, multicenter, observational, longitudinal, open-label, phase IV study conducted between November 2010 and February 2016. The study was conducted in accordance with principles of the Declaration of Helsinki and the protocol, as well as the good clinical practice (ICH-GCP E6) and the applicable national legal and regulatory requirements. The study protocol was approved by the institutional review board at each study site, and written informed consent or assent was obtained from all subjects’ parents or legal guardians before enrolment.

Patients

Subjects (aged 2-18 years or >18 years without fusion of growth plates) who received Saizen (Merck KGaA) via the easypod electromechanical device were enrolled. Subjects who were receiving growth hormones in whom growth plates had fused (ie, for taking growth hormones for its metabolic effects), subjects with contraindications to Saizen as per locally approved prescribing information, subjects using an investigational drug, or subjects participating in an interventional clinical study were excluded from the study. The duration of follow-up for growth hormone treatment was planned to be at least 6 months and up to 5 years. There was 1 baseline visit followed by 1-4 subsequent visits per year as per routine practice. All assessments were performed during the visits. As an observational study, growth hormone treatment and other aspects of patient management were entirely at the discretion of the physician and his or her patient, following a standard clinical practice.

Data Collection and Study Endpoints

The primary endpoint was treatment adherence rate (percentage of prescribed injections that were administered) over time. Data

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on injection time, date, dose, planned frequency were uploaded to a secure web-based database via a specific connection kit and the physician’s computer. For subjects who had consented to participate in the observational study, deidentified data were then uploaded to the web-based registry/observational study. While adherence data from the enrolled subjects were primarily derived from the easyPod device, other information such as demographics, relevant medical and treatment history, and auxological data (eg, height, growth velocity, and bone age) were entered by the physician into the electronic case report form. Data were collected at every visit, as available per routine practice.

Statistical Analysis
Analysis sets included a full analysis set and a complete analysis set. The full analysis set consisted of all the subjects included in the study, whereas the complete analysis set consisted of all the subjects of the full analysis set without missing the treatment start date on the electronic case report form, without gap in the injection information of more than a week after the start of the treatment, and with height measurement closest to the treatment start date not missing using a window of 3 months (91 days). All statistical analyses on adherence rates were performed on the complete analysis set and were performed in a descriptive way for the endpoints, considering this was a single-arm, noninterventional study. Continuous variables were described with the number of subjects, number of subjects with missing data, mean (SD), median, first and third quartiles (Q1, Q3), and minimum and maximum values. For categorical variables, summary statistics were the number and percentage of subjects in each category. To calculate height standard deviation score (SDS) and height velocity (HV) SDS, the reference median growth parameter and the SD of the reference growth parameter were applied. The World Health Organization reference growth table [23] and the Tanner and Whitehouse reference growth table [24] were used for height SDS and HV SDS derivation, respectively.

Results
Patient Characteristics
The ECOS was conducted in 3 medical centers in Taiwan. A total of 35 children had sufficient data and were included in the full analysis set, of which 13 subjects fulfilled the criteria of the complete analysis set. Among the 35 subjects of the full analysis set, 32 had growth hormone deficiency, 2 were born small for gestational age, and 1 had Turner syndrome. The average age was 12.26 years. More than half of the subjects were male (19/35, 54%). At baseline, all subjects were growth hormone–naïve, with a mean height of 137.06 cm and a mean growth velocity of 4.14 cm/year. The baseline demographic characteristics and auxological data are shown in Table 1.

Table 1. Demographic and auxological data of the subjects at baseline.

<table>
<thead>
<tr>
<th>Demographic and auxological data</th>
<th>Full analysis set (N=35)</th>
<th>Complete analysis set (n=13)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years) (min, max)</td>
<td>12.26 (7.0, 16.0)</td>
<td>12.08 (6.0, 16.0)</td>
</tr>
<tr>
<td>Sex, n (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>16 (46)</td>
<td>5 (39)</td>
</tr>
<tr>
<td>Male</td>
<td>19 (54)</td>
<td>8 (61)</td>
</tr>
<tr>
<td>Asian ethnicity, n (%)</td>
<td>35 (100)</td>
<td>13 (100)</td>
</tr>
<tr>
<td>Pubertal stage, n (missing)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tanner 1, n (%)</td>
<td>4 (19)</td>
<td>0</td>
</tr>
<tr>
<td>Tanner &gt;1, n (%)</td>
<td>17 (81)</td>
<td>5 (100)</td>
</tr>
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<td>IGF-1 status, n (missing)</td>
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<td></td>
</tr>
<tr>
<td>Abnormally low, n (%)</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Normal, n (%)</td>
<td>8 (80)</td>
<td>2 (100)</td>
</tr>
<tr>
<td>Abnormally high, n (%)</td>
<td>2 (20)</td>
<td>0</td>
</tr>
<tr>
<td>Bone age, n (missing)</td>
<td>15 (20)</td>
<td>9 (4)</td>
</tr>
<tr>
<td>Greulich and Pyle assessment (years), (min, max)</td>
<td>11.84 (3.0, 15.7)</td>
<td>10.72 (3.0, 15.0)</td>
</tr>
<tr>
<td>Growth velocity (cm/year), (min, max)</td>
<td>4.14 (0.0, 9.4)</td>
<td>3.05 (0.0, 4.9)</td>
</tr>
<tr>
<td>Height (cm), (min, max)</td>
<td>137.06 (103.0, 161.0)</td>
<td>139.26 (103.0, 161.0)</td>
</tr>
<tr>
<td>Indication for growth hormone treatment, n (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Growth hormone deficiency</td>
<td>32 (91)</td>
<td>11 (84)</td>
</tr>
<tr>
<td>Small for gestational age</td>
<td>2 (6)</td>
<td>1 (8)</td>
</tr>
<tr>
<td>Turner syndrome</td>
<td>1 (3)</td>
<td>1 (8)</td>
</tr>
<tr>
<td>Other</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Adjusted mid-parent’s height (cm), (min, max)</td>
<td>162.79 (151.0, 179.0)</td>
<td>164.55 (153.0, 179.0)</td>
</tr>
</tbody>
</table>
Adherence Rates of the Subjects

The primary endpoint was the adherence rate of subjects receiving Saizen via easypod over a period of time. Among the 13 subjects in the complete analysis set, the longest follow-up period was approximately 1.5 years, with a mean (SD) treatment duration of 332 (113.1) days, and the proportions of subjects with adherence data available for 3, 6, 9 months, and 1 year were 100% (13/13), 100% (13/13), 62% (8/13), and 31% (4/13), respectively. The median (IQR) of adherence rates over increasing periods of follow-up are presented in Figure 1. The mean adherence rate was 87.6% at 3 months, 84.3% at 6 months, and 81.0% at 9 months, indicating a slight decrease in adherence rate over time. The mean adherence rate was calculated by averaging all patients’ adherence rates during a period of time. The majority of the complete analysis set subjects maintained an adherence rate of ≥80%, and these percentages remained steady at 3, 6, and 9 months (Figure 2). Subgroup analysis by sex revealed that the median adherence rates were similar between the female and male subjects (Figure 3).

Figure 1. Treatment adherence rates over time (complete analysis set). Boxes show Q1 and Q3, with median as white line and mean as red squares.

Figure 2. The proportion of patients treated with growth hormone using easypod with adherence rates of at least 80% over time and for all patients at any time within the study period.
Growth Outcomes of the Subjects
After 1 year of treatment, subjects had a mean (SD) change in height of 6.25 (3.07) cm, height SDS of +0.27 (0.30), mean (SD) HV of 6.49 (2.95) cm/year, and mean (SD) change in HV SDS of 1.51 (2.22). The growth outcomes and changes from baseline after 1 year of growth hormone treatment are summarized in Table 2. Spearman product-moment correlations between these outcomes and adherence rates were assessed to further investigate the impact of adherence on growth outcomes. Nevertheless, limited by the number of subjects with available data (n=4), no significant and consistent correlation was identified in the complete analysis set (data not shown). Overall, growth outcomes after 1 year indicated a positive growth response to growth hormone treatment.

Table 2. Growth outcomes and changes from baseline of the subjects after 1 year of growth hormone treatment using easypod (complete analysis set).

<table>
<thead>
<tr>
<th>Growth outcome</th>
<th>Subjects with growth hormone deficiency (n=11)</th>
<th>Subject who was small for gestational age (n=1)</th>
<th>Subject with Turner syndrome (n=1)</th>
<th>Overall (n=13)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Baseline height (cm), mean (SD)</td>
<td>141.95 (14.32)</td>
<td>103.00</td>
<td>146.00</td>
<td>139.26 (17.05)</td>
</tr>
<tr>
<td>Change in height (cm), mean (SD)</td>
<td>6.20 (3.24)</td>
<td>8.50</td>
<td>4.50</td>
<td>6.25 (3.07)</td>
</tr>
<tr>
<td>Baseline height, SDS (SD)</td>
<td>–1.74 (0.94)</td>
<td>–2.68</td>
<td>–2.38</td>
<td>–1.86 (0.91)</td>
</tr>
<tr>
<td>Change in height SDS at 1 year, mean (SD)</td>
<td>0.22 (0.29)</td>
<td>0.59</td>
<td>0.56</td>
<td>0.27 (0.30)</td>
</tr>
<tr>
<td>Baseline height velocity (cm/year), mean (SD)</td>
<td>3.30 (1.17)</td>
<td>3.92</td>
<td>0</td>
<td>3.05 (1.47)</td>
</tr>
<tr>
<td>1-year height velocity (cm/year), mean (SD)</td>
<td>6.56 (3.12)</td>
<td>7.92</td>
<td>4.33</td>
<td>6.49 (2.95)</td>
</tr>
<tr>
<td>1-year height velocity SDS, mean (SD)</td>
<td>1.45 (2.31)</td>
<td>2.23</td>
<td>_b</td>
<td>1.51 (2.22)</td>
</tr>
</tbody>
</table>

aSDS: standard deviation score.
bNot available because of missing data.

Discussion
The ECOS assessed the adherence to recombinant human growth hormone treatment as well as growth outcomes in pediatric patients with growth disorders. The results of the European and American countries involved in this open-label, observational, longitudinal study have been published [20-22]. To our knowledge, there is no literature exploring the adherence to growth hormone treatment in Taiwanese or Chinese pediatric patients in a real-life setting, especially by using an electronic monitoring method. In Taiwan, physicians’ clinical experience has shown that some children have relatively low adherence owing to late administration of growth hormones with late bedtime. The culture and living habits of Asians such as children’s daily routines, the time of going to and coming from school, and the activities after school are quite different from others in the world. The medication adherence might be possibly influenced by these differences. In this study, the Taiwanese cohort of the ECOS is reported. The mean adherence rate was generally high in the first year of the treatment, with the majority of the complete analysis set subjects maintaining an adherence rate of greater than 80%. Although the adherence gradually decreased with a longer duration of follow-up, it is in line with the global ECOS results [19] as well as with that of previous studies showing that the adherence rate diminished over time [15,25].
Reduced adherence to growth hormone therapy is detrimental to therapeutic outcomes [11,14,26] and is considered one of the major causes of suboptimal growth [27]. To maximize the effect of growth hormone therapy, it is necessary to maintain good adherence throughout the entire treatment course. Nonadherence not only represents an obstacle to effective treatment, but it also leads to an increase in the medication costs from direct and indirect aspects [13]. Previous literature has shown that adherence could be negatively or positively associated with a variety of factors such as reduced HV [13], comprehensive medical education/training [28], duration of treatment [10,26,29], and choice of injection device [29]. Through real-time monitoring, timely interventions can be prompted in response to nonadherence, rather than signaled by suboptimal growth at a later stage. Moreover, with reliable information regarding adherence at hand, physicians are able to tell whether suboptimal growth arises from nonadherence or other possible causes.

The prevalence and the level of adherence rate vary considerably among studies, which is partly attributable to the methods applied as well as inconsistent definitions used across the studies [13]. While it has been reported that 39%-66% of the patients missed more than 1 injection per week [10,14], another study showed that the median adherence rate might be up to 95% [30]. Most previous studies investigating adherence were cross-sectional, and the adherence was assessed with a questionnaire-based survey [28,29,31]. As an electronic monitoring device is currently recognized as a standard for quantifying adherence [16], it provides information of the precise time and doses of injections, which allows further analyses for nonadherence patterns [16]. A concordance of 84.3% between adherence reported by patients and recorded using easypod has been demonstrated in a study, and the authors found that there was a trend toward self-reported adherence being higher than the recorded adherence [7]. Nevertheless, it is not known whether the difference resulted from forgetfulness, fear of disappointing practitioners, or a combination of factors [32], and no data are currently available to assess this supposition.

In this study, one of the study objectives was to describe the impact of adherence on clinical outcomes for subjects receiving Saizen via easypod. In fact, with 1190 evaluable subjects, the ECOS global results revealed that statistically significant correlations of 0.13 and 0.08 were observed between adherence rate and change in height SDS and between adherence rate and HV SDS, respectively, indicating a positive correlation between adherence rate and growth outcomes. Unfortunately, the number of subjects was not sufficient to support such analyses for the subgroup of Taiwanese patients, since only 4 patients were administered Saizen for more than 1 year. To consolidate the correlation between adherence and growth outcomes, larger sample sizes are required for future studies.

Suboptimal adherence is a common problem in growth hormone treatment. Since adherence to growth hormone therapy is critical for the optimization of treatment outcomes, it has to be taken into account while evaluating the therapeutic effects for treatment modulation in routine clinical practice. Detection of nonadherence can be difficult using pre-electronic monitoring methods because the patient may be reluctant to admit such behavior [7]. The electronic monitoring of injections via devices such as easypod provides reliable and objective information on how many doses have been taken as prescribed and about the nonadministered doses, which reflect the extent to which the patient is adherent to the therapy. The electronic monitoring device, as distinct from conventional monitoring methods, is less labor-intensive and enables physicians to review the timing, date, and dosage of recombinant human growth hormone delivered in a real-time manner. It may help promote adherence and prompt disease management for routine practice.

This study was restricted by its observational nature as there was a considerable level of missing data and intersubject variability. In addition, as mentioned above, the number of subjects included in the complete analysis set was also limited, and most patients had a treatment duration of less than 1 year. Nevertheless, this paper shows the adherence patterns of pediatric patients using an electronic monitoring device, which have not been previously reported in a Taiwanese patient population. To the best of our knowledge, this is the first study providing insight into the adherence rate and characteristics of Taiwanese pediatric patients who require growth hormone treatment. Of note, future studies are warranted to confirm the results and to further explore the effects of individual variables such as bone age at baseline, socioeconomic statuses, and parental, marital, or employment status.

This was a phase IV, open study, and its conditions were different from the phase II or phase III randomized controlled trial. As a phase IV postmarketing study, it generally aims to explore treatment effectiveness and long-term safety. Compared with a randomized controlled trial, observational trials usually reflect the actual clinical treatment effectiveness because the trial design does not have as many limitations in the inclusion/exclusion conditions as a randomized controlled trial. This study did not specify the length of time of patients receiving easypod treatment. Many patients were treated for less than 1 year. In addition to being limited by the number of results, the length of the treatment period is also one of the possible reasons.

Collectively, this study unveils the adherence over time among Taiwanese pediatric patients receiving growth hormone treatment via the easypod electronic monitoring device. The growth outcomes and changes after 1 year of treatment are also presented, although the associations between adherence rate and growth outcome as well as factors affecting adherence to growth hormone therapy in Taiwanese patients were limited by the sample size. The electronic monitoring/injection device serves as a useful tool for both patients and physicians to help disease management and provide direct information regarding adherence to growth hormone therapy.
Acknowledgments

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Conflicts of Interest

PHS, CY, and MCC declare no conflicts of interest. CLC is an employee of Merck Ltd.

References


Abbreviations

ECOS: easypod connect observational study
HV: height velocity
SDS: standard deviation score
Patient-Generated Health Data in Pediatric Asthma: Exploratory Study of Providers' Information Needs

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Abstract

Background: Adolescents are using mobile health apps as a form of self-management to collect data on symptoms, medication adherence, and activity. Adding functionality to an electronic health record (EHR) to accommodate disease-specific patient-generated health data (PGHD) may support clinical care. However, little is known on how to incorporate PGHD in a way that informs care for patients. Pediatric asthma, a prevalent health issue in the United States with 6 million children diagnosed, serves as an exemplar condition to examine information needs related to PGHD.

Objective: In this study we aimed to identify and prioritize asthma care tasks and decisions based on pediatric asthma guidelines and identify types of PGHD that might support the activities associated with the decisions. The purpose of this work is to provide guidance to mobile health app developers and EHR integration.

Methods: We searched the literature for exemplar asthma mobile apps and examined the types of PGHD collected. We identified the information needs associated with each decision in accordance with consensus-based guidelines, assessed the suitability of PGHD to meet those needs, and validated our findings with expert asthma providers.

Results: We mapped guideline-derived information needs to potential PGHD types and found PGHD that may be useful in meeting information needs. Information needs included types of symptoms, symptom triggers, medication adherence, and inhaler technique. Examples of suitable types of PGHD were Asthma Control Test calculations, exposures, and inhaler use. Providers suggested uncontrolled asthma as a place to focus PGHD efforts, indicating that they preferred to review PGHD at the time of the visit.

Conclusions: We identified a manageable list of information requirements derived from clinical guidelines that can be used to guide the design and integration of PGHD into EHRs to support pediatric asthma management and advance mobile health app development. Mobile health app developers should examine PGHD information needs to inform EHR integration efforts.


KEYWORDS

information needs; asthma; symptom management; mobile health; patient-generated health data; pediatrics; adolescents
Introduction

Background
Poorly controlled pediatric asthma continues to be a challenge. Pediatric asthma, the leading chronic disease among children, remains prevalent, and improvements in outcomes have stalled [1]. It is estimated that 6 million children under the age of 18 years in the United States have the chronic airway disease [2]. Despite evidence-based clinical guidelines, suboptimal treatment continues to contribute to a lack of asthma control [3]. Pediatric asthma can be managed with medications and trigger avoidance but requires continuous monitoring to assess control and detect triggers [1,4].

Understanding the complete picture of triggers and symptoms is essential for management, requiring health care providers to perform periodic assessments, adjust treatment plans, and personalize care [5]. However, a lack of objective data from patients means that a provider must depend on patient self-report, known to have reliability challenges, to make clinical decisions [6-8]. A potential solution may be the presentation of relevant patient-generated health data (PGHD) directly in the clinical documentation used by providers as they make decisions. PGHD such as biometric and physical activity, surveys, and health history are data captured electronically by patients outside of the clinic or hospital.

Mobile health (mHealth) technologies offer feasible opportunities to engage adolescents, persons between ages 10 to 19 years, in collecting PGHD [9]. Younger generations in every country are more likely than others to own a phone and are likely to use new technologies [8,10]. Moreover, adolescents engage with their mobile devices even while sick or hospitalized [11]. For pediatric asthma patients, mHealth apps support self-management, and wearable sensors provide ongoing monitoring capabilities [12,13]. The types of data collected from patients using smartphone asthma apps include symptoms, medication adherence, night awakenings, physical activity, and peak-flow expiratory rates [4,5,12,14]. Authors of two studies suggested that collecting the patient’s local environmental data, such as pollen counts, ambient temperature, and humidity, should also be considered [3,4]. When shared during clinical encounters, PGHD have the potential to facilitate assessment, diagnosis, and ongoing patient monitoring [15]. Presenting PGHD within the electronic health record (EHR) is envisioned as an optimal approach so that providers do not need to interrupt their cognitive processes and workflows to navigate between different systems.

Not much is known about which PGHD are of value or how to present PGHD to the providers in the EHR. A recent scoping review showed that EHR integration of PGHD is at an emergent phase; another identified only three asthma apps with the ability to share data with other apps [16,17]. Although many asthma apps exist, only a few of the mHealth technologies developed for childhood asthma have elicited feedback from clinicians [8], and even highly rated apps have not reported integration into clinical workflows [5]. The need for EHR data sharing has been recognized [15], and one study concluded that the introduction of smart-inhaler monitoring data into the EHR might support the development of individualized asthma treatment plans [7].

Despite the potential benefits, clinicians have expressed concerns that incorporating PGHD into the EHR will further contribute to information overload [13,18]. Additionally, studies reported issues with embedding mHealth technologies into clinical workflows and identified uncertainties about organizational readiness to integrate other data sources [13]. To ensure the clinical utility of PGHD, it is vital to understand the clinical workflows in which to integrate PGHD, as well as the specific tasks and decisions that PGHD must support and the relevant information needs of providers. Moreover, the discovery of information needs is necessary to inform future mHealth app implementations.

Purpose
The purpose of this exploratory study was to identify and characterize a discrete set of tasks, decisions, and information needs of providers caring for patients with pediatric asthma and assess whether PGHD might provide useful information. We used outpatient care of patients with pediatric asthma as an exemplar clinical encounter where PGHD might have clinical value. By understanding these needs, we will be able to design interfaces and displays that optimally support the integration of PGHD into EHRs for the management of pediatric asthma.

Methods

Framework and Recruitment
We applied qualitative, descriptive methods to gain insights into key provider tasks, decisions, and information needs regarding PGHD and pediatric asthma. The procedures included analyzing published clinical guidelines to identify relevant decisions and consulting with providers treating patients with pediatric asthma to validate a discrete set of tasks and elicit their perspectives and priorities regarding the decisions, information needs, and potentially relevant PGHD.

We referred to the 3-phase model of needs assessment described by Altschuld and Kumar [19] as a guide. This model or framework proposes a practical process to assess needs that can be molded for a specific situation or setting. In the first phase of the model, preassessment, the goal is to determine what is already known regarding clinician needs and PGHD. We considered the preliminary examination of existing clinical guidelines as the activity to satisfy the preassessment phase or part 1 of this study. For the second phase or part 2 of the study, the assessment consisted of needs assessment procedures and data collection and validation with experts to move toward a full understanding. The third phase of the model, postassessment, involves the identification of strategies or development of solutions to meet the needs that were found during the assessment phase. We will consider the activities related to the postassessment phase in future research.

We recruited a convenience sample of three subject-matter experts (SMEs), domain specialists in pediatric asthma. Although no empirical evidence exists for the most appropriate number of experts for guideline review, similar studies that explored knowledge elicitation for consensus-based guidelines
used at least three task experts [20,21]. We solicited SMEs by reputation using local clinical contacts. Inclusion criteria were the ability to read, understand, and use English as a primary language; self-reported expertise in pediatric asthma; and a history of medical practice in the United States. We excluded providers with adult-only asthma experience, and we did not compensate participants for their time. We received consent from all participants and obtained ethical approval for this study from the institutional review board of the University of Utah.

**Identify Tasks, Decisions, and Patient-Generated Health Data**

We began with a review of the evidence-based pediatric asthma guidelines. We used the two main authoritative sources in pediatric asthma management from the National Institutes of Health National Heart, Lung, and Blood Institute (NHLBI) Expert Panel Report 3: Guidelines for the Diagnosis and Management of Asthma [22] and the Global Initiative for Asthma (GINA) Pocket Guide for Health Professionals [23]. The NHLBI asthma guidelines, in place for more than 25 years, focus on treatment protocols and monitoring for quality asthma care [3]. The GINA report serves as a practical tool to support asthma care and provides the basis for ongoing guideline revisions [24]. The development of these guidelines consisted of formal consensus methods commonly used for clinical guidelines.

According to the preassessment phase of the model, we conducted the needs assessment procedures and derived tasks, decisions, and information needs directly from the guidelines. In this context, a task is a professional duty or clinical responsibility related to patient care [25]. In the development of valid clinical guidelines, tasks are recommended to satisfy the goals of the guideline. Each of the tasks is linked to decisions: cognitive activities involving choices between alternatives or choices about what to believe or what to do [26]. In order to support the appropriate decision, information must be acquired from a person or an external system (an information need). One informatics expert on the research team extracted a list of high-level tasks from the GINA report and the primary task components from the NHLBI guidelines. Most of these were readily identified within each of the guideline documents, with tasks and decisions explicitly identified as such. Then, using the guidelines, the high-level decisions supported by each of the tasks were identified and listed alongside the information collected from the patient that assists with, or could assist with, making the decision. Once the extraction of tasks, decisions, and information needs was competed, the list was discussed with two other clinical informatics experts from the research team for agreement.

To further explain the extraction process, we used the assessment and monitoring task identified in the NHLBI guidelines as an example [22]. The assessment and monitoring task section of the guidelines identified two major decisions: assess the severity of the child’s asthma and decide the level of asthma control. The guidelines listed several information needs related to the decision for severity and control, such as frequency and intensity of the symptoms, functional limitations, exacerbations, lung function, and adverse effects from medication. The information needs were not labeled as such but were obvious from the text of the guidelines. Once we completed this exercise for all tasks from each of the guidelines, we synthesized the findings from both sources to create a single integrated set.

After we assembled the set of tasks, decisions, and information needs, we searched the literature for exemplar asthma mobile apps to assess whether PGHD might provide useful information. In January 2020, we searched PubMed using the terms *asthma mobile health applications* for studies that described asthma PGHD collection features. Given the small number of publications, we did not limit the search to pediatric-specific asthma apps. We examined the types of PGHD collected by each asthma mHealth app [7,14,17]. We inferred the ability of the discovered PGHD types to meet specific information needs by referring to the literature and using our clinical knowledge. Continuing from our previous example, one of the decisions for the assessment and monitoring task is to evaluate the level of asthma control. One asthma app collects the answers from the patient or caregiver and calculates an Asthma Control Test (ACT) score. The ACT is a well-validated, symptom-based tool used to assess symptom control that correlates clinically with specialist ratings and lung function [27]. The ACT is widely used and commonly part of strategies to stratify patients as having poorly controlled or well-controlled asthma [28].

We matched the discovered PGHD types to the corresponding information need in the integrated set. We continued with this process until we had a full set of mapped decisions, information needs, and PGHD for each major task category. All three clinical informatics experts from the research team reviewed the final set of tasks, decisions, information needs, and PGHD types and achieved consensus through discussion.

**Clinician Perspectives**

We scheduled a 30-minute, in-person meeting with each SME independently to review the integrated set of tasks, decisions, information needs, and PGHD types. We also solicited general perceptions of the use of PGHD for adolescent asthma management. In a systematic fashion, we presented the SMEs with the mapped list and asked if it was the right list, if the items were in the order of importance for asthma treatment, and their general thoughts on using PGHD in practice.

We assessed the suitability of PGHD to support their information needs and generated field notes throughout the interview process. Based on the expert feedback, we created a final prioritized list of decisions, information needs, and PGHD types. We recorded participant responses as notes, examined the field notes for themes, and summarized responses. All three clinical informaticists reviewed the findings.

**Results**

**Information Needs and Patient-Generated Health Data Types**

Our analysis of the GINA report and NHLBI guidelines identified 4 high-level tasks:

- Assessment and monitoring

We scheduled a 30-minute, in-person meeting with each SME independently to review the integrated set of tasks, decisions, information needs, and PGHD types. We also solicited general perceptions of the use of PGHD for adolescent asthma management. In a systematic fashion, we presented the SMEs with the mapped list and asked if it was the right list, if the items were in the order of importance for asthma treatment, and their general thoughts on using PGHD in practice.

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• Education for self-management in partnership with the patient and family
• Control of environmental factors and comorbid conditions
• Clinical management and pharmacotherapy

We found that many decisions corresponded to each of the tasks and that some decisions had multiple information needs. This analysis identified several key decisions needed to accomplish the 4 guideline-derived tasks. In our examination of exemplar mobile apps for asthma, we found 9 mHealth apps and 15 different PGHD types (Table 1). We matched the types of PGHD to the information needs derived from the guidelines (Table 2). However, we found that not all types of PGHD configured in the mHealth apps correspond directly with a guideline-derived information need.

Table 1. Asthma mobile health apps and patient-generated health data types.

<table>
<thead>
<tr>
<th>App</th>
<th>Asthma action plan</th>
<th>ACTa</th>
<th>Journal</th>
<th>Activity level</th>
<th>Symptoms</th>
<th>Triggers</th>
<th>Medication reminders</th>
<th>Peak expiratory flow</th>
<th>Inhaler use</th>
<th>Medication use</th>
<th>Environmental factors</th>
<th>Survey data</th>
<th>Location</th>
<th>Mood</th>
</tr>
</thead>
<tbody>
<tr>
<td>Asthma MDb</td>
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<td>Asthma Health Appc</td>
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<td>Asthma Storylinesb</td>
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<td>Kagen Airb</td>
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<td>Kiss My Asthma b</td>
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<tr>
<td>My Asthma Palb</td>
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<td>Smart Trackd</td>
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<td>Propeller Healthb</td>
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</tbody>
</table>

aACT: Asthma Control Test.
bKagan and Garland [17].
cGenes et al [14].
dChan et al [7].

Table 2. Types of asthma-relevant patient-generated health data.

<table>
<thead>
<tr>
<th>Patient-generated health data</th>
<th>Information generated</th>
</tr>
</thead>
<tbody>
<tr>
<td>Asthma Control Test</td>
<td>Symptom trajectory from the last 4 weeks</td>
</tr>
<tr>
<td>Exposures</td>
<td>Symptom triggers such as allergens, smoking, and perfume</td>
</tr>
<tr>
<td>Activity level</td>
<td>Level of physical activity</td>
</tr>
<tr>
<td>Symptoms</td>
<td>Type of symptoms and if daytime or nighttime</td>
</tr>
<tr>
<td>Peak-flow meter</td>
<td>Measurement of peak expiratory flow rates</td>
</tr>
<tr>
<td>Inhaler use</td>
<td>Medication adherence, last dose, missed doses</td>
</tr>
<tr>
<td>Asthma action plan</td>
<td>Progression toward goals and attitudes</td>
</tr>
<tr>
<td>Environmental factors</td>
<td>Pollen count and air quality</td>
</tr>
<tr>
<td>Concerns and/or questions</td>
<td>Ability to recognize worsening symptoms</td>
</tr>
</tbody>
</table>

Perceptions About Patient-Generated Health Data and Pediatric Asthma Management

In August 2019, three primary care providers—two physicians and a nurse practitioner—participated in part 2. Based on their input, including their suggestions on importance to asthma treatment, we modified the initial guideline-derived list; the final list of high-priority information elements is provided in Table 3. The SMEs indicated that there were additional information needs related to triggers of asthma symptoms such as an insufficient level of dustproofing, pets, inadequate pest control measures, cleaning fluids, and other allergens; all of which may not be captured by PGHD. There was a specific interest in pollen, grass, pollution, and other environmental...
Factors, and we added these triggers to the information needs of the decision point on determining exposure to risk factors. Although we identified decisions related to diagnosing in our initial integrated set, we excluded diagnostic decisions from our reviews with the providers based on our assumption that asthma-specific PGHD would be most useful for, and most likely collected by, children already diagnosed with asthma.

Table 3. Guideline-derived decisions and information needs with types of patient-generated health data.

<table>
<thead>
<tr>
<th>Decision</th>
<th>Information needs</th>
<th>PGHDa</th>
</tr>
</thead>
<tbody>
<tr>
<td>Determine level of symptom control</td>
<td>Symptom trajectory, types of symptoms, medication adherence, last dose, missed doses</td>
<td>Symptoms, ACTb, inhaler use</td>
</tr>
<tr>
<td>Determine exposure to risk factors</td>
<td>Symptom triggers such as allergens, smoking, pollen, poor air quality, perfume, inadequate dustproofing, pets, inadequate pest control measures, and cleaning fluids</td>
<td>Exposures, symptoms, environmental factors</td>
</tr>
<tr>
<td>Determine adjustments to medication regimen</td>
<td>Symptom trajectory, medication adherence, last dose, missed doses</td>
<td>Symptoms, inhaler use</td>
</tr>
<tr>
<td>Determine adjustments to action plan</td>
<td>Progression toward goals, attitudes, child’s and family’s ability to recognize worsening symptoms, level of physical activity</td>
<td>Asthma action plan, activity level</td>
</tr>
<tr>
<td>Determine ability to take medication</td>
<td>Observation of inhaler technique, medication adherence, last dose, missed doses</td>
<td>Inhaler use</td>
</tr>
<tr>
<td>Determine lung function</td>
<td>Lung function assessment, peak flow expiratory rates</td>
<td>Peak flow meter</td>
</tr>
<tr>
<td>Determine educational needs</td>
<td>Subjective questions or concerns from child or family</td>
<td>Concerns and/or questions</td>
</tr>
</tbody>
</table>

aPGHD: patient-generated health data.
bACT: Asthma Control Test.

A few common perspectives resulted from the input of the SMEs. Each placed primary focus on uncontrolled asthma and indicated that PGHD would be most useful for adolescents, a subset of pediatric patients, who have trouble controlling their asthma. Perspectives concerning the timing for viewing PGHD were also prevalent. The SMEs expressed a desire to see the PGHD in the EHR at the time of the visit. They thought that it would be unusual to view the PGHD before a patient visit or between visits without an alert in the EHR or communication from the patient. Last, there was an interest in observing the inhaler technique and knowing whether the patients use spacers with their inhalers. The SMEs viewed proper inhaler technique as a critical component of medication adherence and pediatric asthma self-management. They thought that although patients might perform the proper inhaler technique during clinic visits, the technique might be inadequate outside of the visits.

Discussion

Principal Findings

A complete understanding of the information needs supported by PGHD is essential for the seamless integration of PGHD into workflows [13]. Identification of provider information needs is the first step in supporting the integration of PGHD into EHRs. In this study, we identified a list of high-priority decisions, information needs, and potential PGHD sources that can address the information needs of providers treating patients with pediatric asthma. We believe our findings demonstrate the suitability of PGHD to support clinical decisions for pediatric asthma. This work serves as a foundation to support future postassessment work such as evaluating the use of PGHD from mHealth apps and the integration of PGHD to EHRs.

In addition to the main findings, we uncovered aspects of PGHD use in pediatric asthma that may inform future research questions. Providers treating patients with pediatric asthma considered the use of PGHD to determine asthma triggers to be an essential part of treatment. Similar to other studies, providers expressed a need to know about general environmental factors, including air pollution and pollen levels [29,30]. We found that providers also wanted to learn about pest control (eg, roaches or rodents) in addition to contact with pets and other animals. Exposure products such as cleaning fluids, detergents, and perfumes were also of interest. Although it may prove difficult to capture all triggers using mobile technologies or sensors, the majority of asthma mHealth apps have the potential to include local air quality [17]. Further exploration is needed to fully understand the clinical utility of the inclusion of triggers in mHealth asthma apps.

The providers commented that inhaler use was an essential gap in their knowledge of patient behaviors, and in our review of the guidelines we found inhaler use to be a clear information need. Previous research reported that many patients use their inhaler poorly or share inhalers with friends or family members [1,8]. In our limited search, we found a lack of smart-inhaler mHealth apps that can capture and transmit data on inhaler technique or the use of spacers to providers. Technologies such as Respiro and Capmedic provide technique-related feedback to the user, but it is unclear whether providers can access technique assessment data [31,32]. As technologies advance, evidence related to the features of audio and video capture of inhaler use may be beneficial.

We found that providers were most interested in PGHD collected by patients whose asthma was severely uncontrolled. According to pediatric asthma guidelines, asthma severity is classified as
mild, moderate, or severe, with severe asthma requiring the highest level of treatment [33]. Given the potential long-term repercussions, it is vital to treat children adequately in order to establish control early in life [34]. Although an emphasis on the PGHD of patients with severe asthma is reasonable, mHealth technologies identified gaps by noncompliant patients [8]. However, a study by Chan et al [7] reported that the use of mobile technologies for severe asthma might be most promising. As the number of mHealth apps increases, it may be worthwhile to collect additional evidence on the PGHD use—or lack of use—of pediatric mHealth apps for all types of asthma severity before focusing solely on uncontrolled cases.

Although particular care models determine the point in the care process when providers should review PGHD, we determined that in the context of outpatient pediatric asthma care, providers preferred to see the PGHD at the time of the visit. Other researchers have described programs that use nurse care coordinators or community health workers to review PGHD on a more ongoing basis [35,36]. Given that many patients with asthma have visits at the time of an exacerbation [1], it is reasonable that specialists in pediatric asthma see the most value for PGHD as part of their during-visit workflows [17].

In this study, providers expressed a keen interest in viewing PGHD directly from the EHR and not from another app. However, there is a risk that the potential richness of PGHD may get lost once it is added to an already complex and sometimes unsearchable EHR. Because of these comments, it would be worthwhile to investigate the requirements and provider preferences for the display of PGHD alongside EHR data and the locations in the EHR that may be most beneficial.

If the future of health care is personalization and individualized approaches to care, new strategies to harvest data from mobile technologies are needed [1]. As a first step, information technology specialists and health care providers should work together to determine clinical information needs for available PGHD and to update needs as new PGHD sources become available. There is great potential for PGHD to support the longitudinal care of patients with chronic disease. An understanding of the PGHD needs for pediatric asthma provides the opportunity to similarly explore the PGHD needs of other chronic diseases. The next step in the needs assessment framework (phase 3) indicates that actions must resolve the needs-based priorities [19]. We suggest a strategy for future research that examines the PGHD visualization and display preferences of providers to support the design of EHR integration.

Limitations
Although this work was grounded in widely accepted guidelines, there may be nuances that were not accounted for, and all providers may not agree with or use the guidelines in practice. In addition, we validated our findings with primary care providers. It may be helpful to explore information needs with providers in other settings as needed.

Conclusion
To optimally inform implementation approaches that integrate PGHD, the identification of provider information needs is essential. We extracted a set of tasks, decisions, and information needs derived from clinical guidelines and aligned them with PGHD types that may be collected by patients. By reviewing with providers caring for pediatric asthma patients in the outpatient setting, we validated the information needs and found that they align with some types of PGHD currently collected. This preliminary work serves to support the future design and development of mHealth apps and methods to integrate PGHD into EHRs that are in alignment with clinical information needs for chronic disease management.

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Conflicts of Interest
None declared.

References

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Abbreviations

ACT: Asthma Control Test
EHR: electronic health record
GINA: Global Initiative for Asthma
mHealth: mobile health
NHLBI: National Heart, Lung, and Blood Institute
PGHD: patient-generated health data
SME: subject-matter expert

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