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Development of a mobile application based on the salutogenic model for self-management in adolescents with Moyamoya disease

Won-Oak Oh, RN, PhD [a,1], Il Tae Park, RN, PhD [b,2], Jihee Han, RN, MSN [a,1], Eunji Lee, RN, BSN [a,1], Anna Lee, RN, PhD [c,3,*]

[a] Korea University, College of Nursing, Seoul, Republic of Korea
[b] Woosong University, College of Health and Welfare, Daejeon, Republic of Korea
[c] Yonsei University, College of Nursing and Mo-Im Kim Nursing Research Institute, Seoul, Republic of Korea

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ABSTRACT

Purpose: Moyamoya disease (MMD) is a rare disease which has a high incidence of onset in adolescence. Disease self-management skills are imperative for adolescents with MMD. This study aimed to describe the systematic development, content, and usability of the Moyamoya Healthy Youth application (app), which was developed to enhance self-management skills for adolescents with MMD.

Design and methods: The theoretical grounding for the app was salutogenic model and the development process of the app was guided by the intervention mapping (IM) protocol. Results of each IM step were applied to the next step leading to the design of the app. Additionally, a pilot test was conducted to determine the usability of the app.

Results: Following the salutogenic model, we identified the stressors, behaviors, and resources regarding managing symptoms of MMD by interviewing adolescents with MMD, their parents, and healthcare providers. Based on the findings of the interviews, we determined the program outcomes and performance objectives to improve the self-management of MMD in adolescents. The app was developed by translating the theoretical methods to achieve the performance objectives into practical strategies for delivering the program. A pilot test with eight participants showed satisfaction with the app in terms of its usefulness and ease of use.

Conclusion: We delineated the development process of the Moyamoya Healthy Youth. Additionally, we presented the positive outcomes regarding the usability of the app.

Practice implications: The Moyamoya Healthy Youth app could benefit adolescents with MMD, by improving their self-management skills which are crucial for their health.

Introduction

Moyamoya disease (MMD) is a rare, chronic, and progressive intracranial arteriopathy that is often reported in East Asian countries, including South Korea (hereinafter Korea) (Singh et al., 2022). In Korea, the prevalence of MMD has increased from 6.3 per 100,000 people in 2004 to 18.1 in 2015 (Kim, 2016). A trend in rising incidence rates has been observed worldwide (Huang et al., 2017; Lee et al., 2020).

Considering that the onset of MMD is mostly during adolescence and middle age (i.e., 50s), keen attention to these age groups is required (Kim, 2016; Sun et al., 2021). In particular, adolescence is a critical period of dramatic changes in physical, emotional, and social development, as well as the time when one begins establishing their own identity (e.g., body image) and health behaviors (Steinberg, 2019).

Adolescents with MMD are more likely to experience repetitive transient ischemic attacks (TIAs) caused by stenosis or occlusion of the cerebral blood vessels, rather than the cerebral infarction and hemorrhage often exhibited by adults with the disease (Kleinloog et al., 2012). The manifestation of a TIA can be through various symptoms such as seizures, paralysis, and speech, vision, or memory impairments, which can be temporary or permanent (Bang et al., 2016). TIAs have been reportedly triggered by daily activities, such as singing, crying, or

* Corresponding author.
E-mail addresses: wooh@korea.ac.kr (W.-O. Oh), eunjilee0121@korea.ac.kr (E. Lee), annalee@yuhs.ac (A. Lee).
[1] College of Nursing, Korea University, 145 Anam-ro, Seongbuk-gu, Seoul 02841, Republic of Korea.
[2] College of Health and Welfare, Woosong University, 171 Dongdaejeon-ro Dong-gu, Daejeon 34606, Republic of Korea.
[3] College of Nursing, Yonsei University, Mo-Im Kim Nursing Research Institute, Yonsei-ro 50, Seodaemun-gu, Seoul 03722, Republic of Korea.

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excessive exercise, as well as feeling stressed (Kim, 2016; Lee et al., 2017). Potentially harmful conditions often experienced in daily life, such as dehydration and stress, have an influence on cerebral blood flow, which can lead to neurological defects among adolescents with MMD (Hwang, 2010). It is, therefore, vital that adolescents with MMD are equipped with a full understanding of the risk factors for symptoms and the maintenance of physical and emotional health in their everyday lives. Furthermore, enhancing self-management skills to mitigate the risk factors becomes crucial. For example, managing stress, and monitoring activities and eating habits are critical to preventing a reduction in cerebral blood flow.

To date, the cause and cure of MMD have not been thoroughly identified; surgery is regarded as the only major intervention for MMD that purports to alleviate the symptoms, although is not a perfect cure (Takanashi, 2011). This finding also strengthens the importance of self-managed healthcare in daily life among adolescents with MMD.

Using mobile health (mHealth) application (app) technology is promising for improving the self-management of adolescents with MMD. Apps have many advantages, including interactive methods (e.g., animations), constant accessibility, anonymity, possible adjustment to the needs of the users, and tailored feedback (Jeminiwa et al., 2019). Contemporary adolescents, so-called digital natives, are very natural and comfortable with exploring mobile devices and apps (Niksch, 2015). Using mHealth apps is effective in enhancing adolescents' engagement in health behaviors across various chronic conditions, including diabetes, asthma, and mental health (Badawy et al., 2017; Cafazzo et al., 2012; Hammonds et al., 2015; Ostojic et al., 2005).

As approximately 95% of Korean adolescents reportedly have smartphones (Korean Media Panel Survey, 2020), Korea is known to be a highly wired country. Interventions using mHealth are expected to have positive effects on Korean adolescents with MMD. Accordingly, we decided to develop a mobile app, the Moyamoya Healthy Youth app, to enhance successful management among adolescents with MMD.

The theoretical grounding for our app was the salutogenic model (Antonovsky, 1979). Salutogenesis emphasizes one's resources and abilities to handle stress, rather than focusing on the stress itself, in order to improve one's health (Antonovsky, 1979). In this model, generalized resistance resources (GRRs) refer to individual resources that enhance health, facilitate dealing with stressors, and consist of four aspects: cognitive, interpersonal/relational, emotional, and macro-social cultural aspects (Antonovsky, 1979). Antonovsky (1979) stated that the GRRs contributed to life experiences that featured consistency and the participation of self in shaping outcomes; the GRRs also contributed to the development a sense of coherence (SoC). The SoC refers to an individual's ability to figure out and mobilize resources and consists of three components: comprehensibility, manageability, and meaningfulness (Antonovsky, 1979). The salutogenic model posits that individual life experiences shape one's SoC; a strong SoC encourages one to utilize resources to cope with stressors and successfully manage tension (Antonovsky, 1979). We employed these core concepts of the salutogenic model to develop the mHealth program.

Adolescents with MMD face stressors and tension from various changes in puberty as a typical developmental process, as well as from the management of MMD. It is practical for them to focus on improving their capability to successfully manage their stressors rather than focus on eliminating the disease. MMD is chronic disease that needs to be managed within one's daily life. To enhance their capability for successful management, the resources surrounding adolescents with MMD (GRRs) should be increased (e.g., individual cognitive and emotional capability, peer, or family support). Consequently, these GRRs could shape their experience of MMD which would in turn contribute to their SoC, namely, their ability to identify and utilize the resources to deal with stressors related to MMD. The current app was designed to improve the GRRs and SoC of adolescents with MMD, to enable them to successfully manage their symptoms. The present article aimed to describe the systematic development and content of the Moyamoya Healthy Youth app and establish its usability.

Methods

The development process of the Moyamoya Healthy Youth app was guided by the intervention mapping (IM) protocol (Bartholomew et al., 2016). The IM protocol has been used for theory- and evidence-based health promotion program planning (Bartholomew et al., 2016). Bartholomew et al. (2016) suggested the following six steps: (1) generate a logic model of the problem based on a needs assessment, (2) establish a logic model of change with expected behavioral and environmental outcomes, (3) determine the program design, (4) produce the programs, (5) develop a program implementation plan, and (6) specify the program evaluation plan. Each step requires a defined end task that leads to the developmental activities for the subsequent step. In addition, the mHealth evidence reporting and assessment (mERA) checklist (Agarwal et al., 2016) was taken as a guide to elaborate on the app development results effectively.

The development of the Moyamoya Healthy Youth app was reviewed by the Office of Human Research Ethics at [Authors blinded] University and was determined to be exempt from ethical approval.

Step 1. Logic model of the problem

For the first step of the IM protocol, a problem analysis was conducted for the construction of a logic model (Bartholomew et al., 2016). The core tasks of this step included carrying out a needs assessment and stating the program goals and context (Bartholomew et al., 2016).

To identify the problems and needs experienced by adolescents with MMD, we reviewed prior studies on MMD. The reviewed studies included empirical studies or clinical guidelines that targeted adolescents with MMD and were published within the last 20 years. Additionally, we conducted both individual interviews and a focus group discussion with adolescents with MMD ($n = 12$), their parents ($n = 11$), and attending healthcare professionals ($n = 8$) to gain an in-depth understanding of their experience, especially regarding symptom management in the period between 4 July 2019 to 10 October 2019. The interview questions were generated based on the salutogenic model (Antonovsky, 1979). Data were analyzed using a deductive content approach. We have provided a detailed account of our findings in our previous study (in press).

Step 2. Program outcomes, objectives, and logic model change

The second step of IM involved setting program goals and outcomes as well as logic model changes regarding the determinants of behavioral and environmental outcomes (Bartholomew et al., 2016). By combining the results from Step 1 and the key aspects of Antonovsky's salutogenic model, we determined detailed program outcomes and objectives that represent behaviors that need to be changed to fulfill the overall goal of the Moyamoya Healthy Youth program, namely, successful self-management among adolescents with MMD (Table 1).

Step 3. Program design

The third step of the IM process was to select theoretical methods, including program themes, theory- and evidence-based change methods, and practical applications to deliver change methods (Bartholomew et al., 2016). Our research team chose theoretical methods for each behavioral determinant in conjunction with the change objectives, which were based on the salutogenic model. We referred to the literature on adolescent self-management or healthy behavioral changes to seek appropriate theoretical methods. Additionally,

Table 1
Performance objectives and relevant determinants pertaining to program outcomes.

Outcome	Performance objective	Determinant	Moyamoya healthy youth
Adolescents with Moyamoya disease increase their **generalized resistance resources**	**PO 1** Improving the **cognitive** aspect of the generalized resistance resources	**Knowledge** The trajectory of MMD Symptom/symptom management	**Resources** *MMD guideline* *Stress management*
	PO 2 Improving the **interpersonal, relational** aspects of the generalized resistance resources	**Positive relationship** Supportive bonding with parents Positive relationship with peers Positive relationship with teachers	**Resources** *Recognizing my strengths* *Facilitating friendship*
	PO 3 Improving the **emotional** aspect of the generalized resistance resources	**Acceptance** Positive acceptance/attitude of one's disease conditions	**Resources** *Recognizing my strengths* **Cheer Up** *Pop up messages*
	PO 4 Improving the **macrosociocultural** aspect of the generalized resistance resources	**Support** Support of health care professionals	**Administrator's management by reviewing health information**
Adolescents with Moyamoya disease increase their **sense of coherence**	**PO 5** Improving the **comprehensibility** aspect of the sense of coherence	**Comprehensibility** Understanding the patterns of TIA symptoms Identification of the signs of TIA symptoms Predicting TIA occurrence of symptoms	**Resources** *MMD guideline*
	PO 6 Improving the **manageability** aspect of the sense of coherence	**Manageability** Developing one's own strength for coping with TIA symptoms Making a social shield for emergencies Diet management Hyperventilation prevention Normalization of daily life Coping with TIA symptoms properly	**Resources** *MMD guideline* *Recognizing my strengths* **Dairy & Daily report** *Exercise* *Medicine* *Sleep* *Food* *Symptom*
	PO 7 Improving the **meaningfulness** aspect of the sense of coherence of adolescents with MMD	**Meaningfulness** Pursuing valuable purpose through disease experience The establishment of the direction of life	**Resources** *Recognizing my strengths*

the results of the interviews conducted as part of the program development process led to the selection of appropriate methods to achieve the program objectives. In terms of the program delivery methods, we decided to employ the mHealth app. The app was developed by a research team consisting of healthcare professionals ($n = 6$) and app development experts ($n = 3$).

Step 4. Program production

The fourth step of the IM process required refinement of the program using specific structures and materials (Bartholomew et al., 2016). We combined the findings and strategies from the literature review and interviews, based on the salutogenic model. The app's contents and program sequences were developed to target the enhanced GRRs and SoC among adolescents with MMD. We created the app with a director for planning the app, an app programmer, and a user interface designer. Additionally, an animator, a professional voice actor, and a yoga instructor were involved in generating specific content for the app. The experts involved in this project verified the appropriateness of the app's contents and methods for the intended population.

Step 5. Program implementation plan

The penultimate step of the IM process focused on planning the implementation and continuation of the program (Bartholomew et al., 2016). Planning involved determining who would implement it and how the program would be conveyed. In this step, we investigated the usability of the app developed through an IM protocol by implementing a pilot intervention study with eight participants (two neurosurgery nurse practitioners, two nursing school faculty members, two experts in app development, and two high school students). We recruited participants by convenience sampling. After the authors explained the purpose of this study to the subjects and obtained their consent, they voluntarily participated in this study. Participants downloaded the Moyamoya Healthy Youth app on their own cell phones from the Google Play Store and used the app for one week.

Step 6. Evaluation plan

The final step of IM involved the evaluation of the developed program including deciding the indicators and measures for assessment (Bartholomew et al., 2016). We evaluated the usability of the app based on technology acceptance model (TAM) (Davis, 1989) which has been used extensively in studies regarding technology acceptance (Atack et al., 2010; Verkuyl et al., 2016). This model posits that perceived ease of use and perceived usefulness are core factors to determine users' acceptance and eventual use of a certain technology (Davis, 1989).

We modified 15 questions developed in previous studies based on the TAM (Atack et al., 2010; Verkuyl et al., 2016) to assess the user's perceived usefulness (6 questions) and perceived ease of use of the app (9 questions). The questions were evaluated using a five point Likert scale. The eight participants who used the Moyamoya Healthy Youth app were asked to complete the questions after using the app for one week. Their responses were analyzed using SPSS 24.0 for descriptive statistics.

Results

Step 1. Logic model of the problem

Individual and focus group interviews with adolescents with MMD, their parents, and relevant professionals yielded crucial insights into the adolescents' experiences from the point of view of salutogenesis. They articulated various stressors caused by internal and external stimuli, including feelings of guilt about their misbehavior that could worsen

their symptoms, especially when facing situations that can cause TIAs (e.g., eating spicy food), relationship difficulties with their overprotective parents, and issues related to school life (e.g., academic accomplishments and maintaining friendships). These challenges can lead to tension, such as fear of incompetence in managing TIAs and worries related to alienation from friends. They also showed concerns about embarrassment related to sudden onset TIAs and conflicts with their parents related to symptom management.

The interviewees stated the critical resources for managing their conditions. Knowledge and information regarding MMD symptoms and relevant management served as a protective factor for adolescents with MMD and their families. Various types of support, including support from parents, peers, and healthcare professionals, were found to be crucial resources to attenuate the challenges of adolescents with MMD. In addition, the interviewees expressed positive acceptance, and that their attitude toward MMD was helpful in managing their symptoms. They noted various factors that influenced their SoC associated with their condition management behaviors. They revealed that understanding the pattern of TIA symptoms and predicting TIA occurrence were helpful in enhancing their management abilities. They also pointed out the importance of developing their own coping strategies to handle their symptoms and establishing social and school support systems for managing MMD. Through the interviews, the pursuit and establishment of worth and purpose in life despite disease experiences was found to be crucial for adolescents with MMD. The interview findings formed the basis of the app's contents.

Step 2. Program outcomes, objectives, and logic model change

Based on the results of the problem analysis conducted in Step 1, we specified the program outcomes and performance objectives. The program outcomes showed that adolescents with MMD enhanced their GRRs and SoC. To achieve the expected outcomes, we stipulated the performance objectives. The performance objectives were also derived from the results of the interviews, in combination with the salutogenesis model (Table 1).

Step 3. Program design

We developed the Moyamoya Healthy Youth app to deliver the program involving the specific program outcomes and performance objectives resulting from Step 2 (Fig. 1). In this section, we addressed the selected theoretical methods and relevant determinants as well as their translation into practical strategies for the performance objectives. Additionally, we verified all the contents of the symptom management in the Moyamoya Healthy Youth app abided by MMD management guidelines provided by the Korea Disease Control and Prevention Agency.

Performance objective 1 - Improving the cognitive aspects of GRRs

We set improvement in the cognitive aspects of GRRs among adolescents with MMD as the performance objective. Previous research focusing on healthy behavioral changes in adolescents states that providing in-depth knowledge and information could be a suitable theoretical method to induce behavioral changes (Brunet et al., 2018; Scheerman et al., 2018). We reflected on the theoretical methods, that is, knowledge regarding MMD and management, in the development of the app.

The Moyamoya Healthy Youth app contains a resources section that includes educational content regarding MMD management guidelines and stress management. The MMD management guidelines include the disease's trajectory, cause, symptoms, progress, and management. The video clips on stress management consist of stretching exercises and stress management strategies for adolescents with MMD (Fig. 1). For the stretching exercises, a professional yoga instructor demonstrates appropriate yoga poses for adolescents with MMD for approximately 13 min. The poses were validated by a neurologist and a neurology nurse practitioner. We also inserted a narration that wa participants to stop the exercise if they experience dizziness or symptoms. The video clips about stress management strateg (e.g., behavior management, exercise, relaxation, and nutrition to duce stress) total approximately seven minutes.

Performance objective 2 - Improving the interpersonal relationship aspe of GRRs

We aimed to enhance the relationship aspects of GRRs amo adolescents with MMD by intervening in their perceptions of self a relationships with others. We determined that improving the positi self-concept of adolescents is crucial for building positive relationshi with others, based on the previous findings of significant associatio between positive self-concept and relationships with others (Hin et al., 2001).

"Recognizing my strength" in the resources section of the app hel to build adolescents' positive aspects of themselves, including their a complishments, strengths, and the feeling of a promising future. Th section attempts to affirm that they are special and valuable peopl This section also enables adolescents to recognize the support systen around them. The video clip intends to highlight the importance of th relationships between adolescents with MMD and their familie peers, or teachers, which could play a vital role in helping adolescen manage their symptoms. Additionally, "facilitating friendship" in the re sources section of the app demonstrates useful skills that can help the make friends and maintain friendships, including communication skill complementing others, asking for help, and saying no to inappropriat requests (Fig. 1).

Performance objective 3 - Improving the emotional aspects of GRRs

We found that adolescents' acceptance of themselves is essential fo improving the emotional aspects of their resources. The "recognizin my strengths" section was created to enhance emotional competenc as a translated strategy in the app. This video clip guides adolescent with MMD to remember their accomplishments and strengths. Th app also has the feature of sending random text messages as promp cues. The messages are intended to provide encouragement for adoles cents with MMD (example messages: "You are not alone," and "You ar such a great person!") (Fig. 1).

Performance objective 4 - Improving the macro-sociocultural aspects o GRRs

We identified continuous support from healthcare professionals a an effective method for improving the macro-sociocultural aspects o adolescent resources. One of the app managers who is a neurology nurse practitioner reviews the health information that app users report every day. If an app user shows an unfavorable or concerning health status based on the reported health information, the manager contacts them to provide emotional support and education depending on their individual needs. By doing this, we intended to enable the person to continue using the app for self-management.

Performance objective 5 - Improving the comprehensibility aspect of the SoC

We ensured that the resources section of the app provides information regarding TIA symptoms in an effort to improve the comprehensibility of adolescents with MMD. This information includes pathophysiology and MMD symptoms.

Performance objective 6 - Improving the manageability aspect of the SoC

To improve the manageability of adolescents with MMD, we found that the development of their strengths for coping with TIA symptoms and emergencies is crucial. The resources section of the app includes contents that facilitate improved self-esteem of adolescents with MMD to develop their strengths and manage their symptoms. The

Fig. 1. Moyamoya Healthy Youth application.

resources section provides specific instructions regarding coping with emergencies.

Moreover, to enhance the adolescents' manageability, we employed goal setting and self-monitoring methods (Ryan & Sawin, 2009). We generated the diary section of the app. Initially, app users input information on their weight to calculate the proper amount of sleeping hours, water intake, and exercise required per day. Then, app users can use the diary section to record their health information, including the amount of exercise, sleeping hours, water intake, medication, and food consumption per day. By helping app users record their health information every day, we intend to enhance their self-management skills.

In terms of exercise, app users are required to report the amount of time spent exercising and the intensity of the exercises, as well as step counts. Exercise intensity was evaluated using the Borg Rating of Perceived Exertion scale with responses ranging from 6 (never hard) to 20 (extremely hard) (Williams, 2017). The amount of water intake is reported by the number of glasses of water consumed per day. The Morisky Medication Adherence Scale (Morisky et al., 1986) is used to report the medication consumption. The scale includes four items with yes (0) or no (1) responses to the following questions: (1) Do you ever forget to take your medicine? (2) Are you careless about taking your medicine at times? (3) When you feel better, do you sometimes stop taking your medicine?, and (4) If you feel worse when you take your medicine, do you sometimes stop taking it? Higher scores indicate better medication adherence (Morisky et al., 1986). For food intake, we developed a checklist including 18 items based on dietary recommendations for adolescents provided by the government (Moon et al., 2014). Finally, users can check whether they experience any of the eight TIA symptoms identified in the previous study per day (in press). In addition, they can input descriptions of their symptoms. Users were required to report their feelings of discomfort using 10 items established in previous studies.

In the daily report section, the users' health information stored in the diary section is visualized using line graphs. If their self-management status reaches their goals, smiley face icons appear on the app screen. Otherwise, sad face icons appear. Users can easily check their information over a two-week period at a glance by manipulating the graphs on the screen (Fig. 2).

Performance objective 7 - Improving the mindfulness aspect of the SOC

The resources section of the Moyamoya Healthy Youth app provides content related to positive perceptions of themselves and their lives as an avenue for improving the meaningfulness of adolescents' lives with MMD. This content helps adolescents realize the meaningfulness of themselves and their lives.

Step 4. Program production

The structure of the Moyamoya Healthy Youth app comprises a server/client system using a hypertext transfer protocol and Web application programming interface (API). The server configuration used the MySQL database and Apache Tomcat/Spring Framework. In order to enhance users' acceptance, this app was designed by a user interface designer, taking into account the youth's preferences.

The client configuration was performed by an Android app using the Android SDK platform. This app was created by setting the API level to 24–29. The server linked to the app was produced using the Spring Boot 2.3.0, Java JDK 11 platform. It took 4 months, from August 2020 to November 2020, for the app development. Servers and databases are strictly secured with firewalls and SSL access. HTTPS is applied for web security, and thorough protection is used to prevent SQL injection, cross-site scripting (XSS), and cross-site request forgery (CSRF), which could be vulnerable in applications. Approximately, $ 30,000 cost for the app development. The app development was financially supported by the Korean government as a part of a government project for rare disease management. This app is freely open to MMD society and associate hospitals.

This app consists of four main components: (1) diary, (2) daily report, (3) resources, and (4) cheer-up messages. Additionally, the app contents included ID creation, login, setting, alarming, and surveys (Fig. 3). First, the user needs to create an ID and fill out the user's information section, including weight. Based on the user's weight, goals of proper sleeping hours and water consumption are determined. The app manager can keep track of how a participant uses the app. At present, the Moyamoya Healthy Youth app can be downloaded as a beta version from the Google play store.

Step 5. Program implementation plan

Eight participants used the Moyamoya Healthy app for one week. They downloaded the app to their smartphones and created an ID. Then, they inputted their information, including weight, which was used to calculate the appropriate amount of water consumption and sleeping hours. Participants recorded their daily exercise hours, sleeping time, dietary and water intake, medication intake, and symptoms using the app diary. The app administrator checked their daily information and any errors.

Step 6. Evaluation plan

The app users perceived the Moyamoya Healthy Youth app as easy to use. The highest mean obtained among the items of the app's ease of use was the adequacy of actuation speed ($M = 5.0$, $SD = 0.00$), followed by the ease of using methods ($M = 4.5$, $SD = 0.75$) (Table 2). The mean scores of clarity and readability of the app's content texts, and the clarity of the tasks' descriptions were 4.38 ($SD = 0.91$, 0.74, and 0.74). Additionally, the mean scores of the screen quality, technical errors, and enjoyment of using the app were 4.25 ($SD = 0.71$), 4.00 ($SD = 0.93$), and 4.00 ($SD = 1.07$), respectively.

The app users showed relatively high levels of perception of the app's usefulness. The highest mean obtained among the app's usefulness domains was helpfulness for stress management (mean [M] = 4.75, standard deviation [SD] = 0.46). The average scores of the app's usefulness in improving MMD knowledge, healthy behaviors, and quality of life were all 4.63 ($SD = 0.52$). In addition, the mean scores of the app's usefulness for enhancing peer relationships and self-esteem were all 4.25 ($SD = 0.89$).

Discussion

In this paper, we describe the development and content of the Moyamoya Healthy Youth app. We intended to induce successful self-management skills in adolescents with MMD by using the Moyamoya Healthy Youth app. A notable strength of the app development is its theoretical grounding, the salutogenic model. The use of the salutogenic model ensured the factors for achieving changes through the Moyamoya Healthy Youth app.

In addition, the IM protocol was used as a guide for the systematic development of the app. The IM protocol has proven to be a useful method for developing health promotion programs. Following the IM process is a time-consuming task, as other researchers experiencing the IM process often point out (O'Connor et al., 2018; Scheerman et al., 2018). Nonetheless, using the IM protocol ensured critical program goals that should be addressed in the Moyamoya Healthy Youth app.

Overall, the participants who participated in this pilot intervention study showed satisfaction with the app in terms of its usefulness and ease of use. The participants expressed that "'stress management" in the resources section was the most useful. Managing stress is one of the critical self-management skills for adolescents with MMD, in that

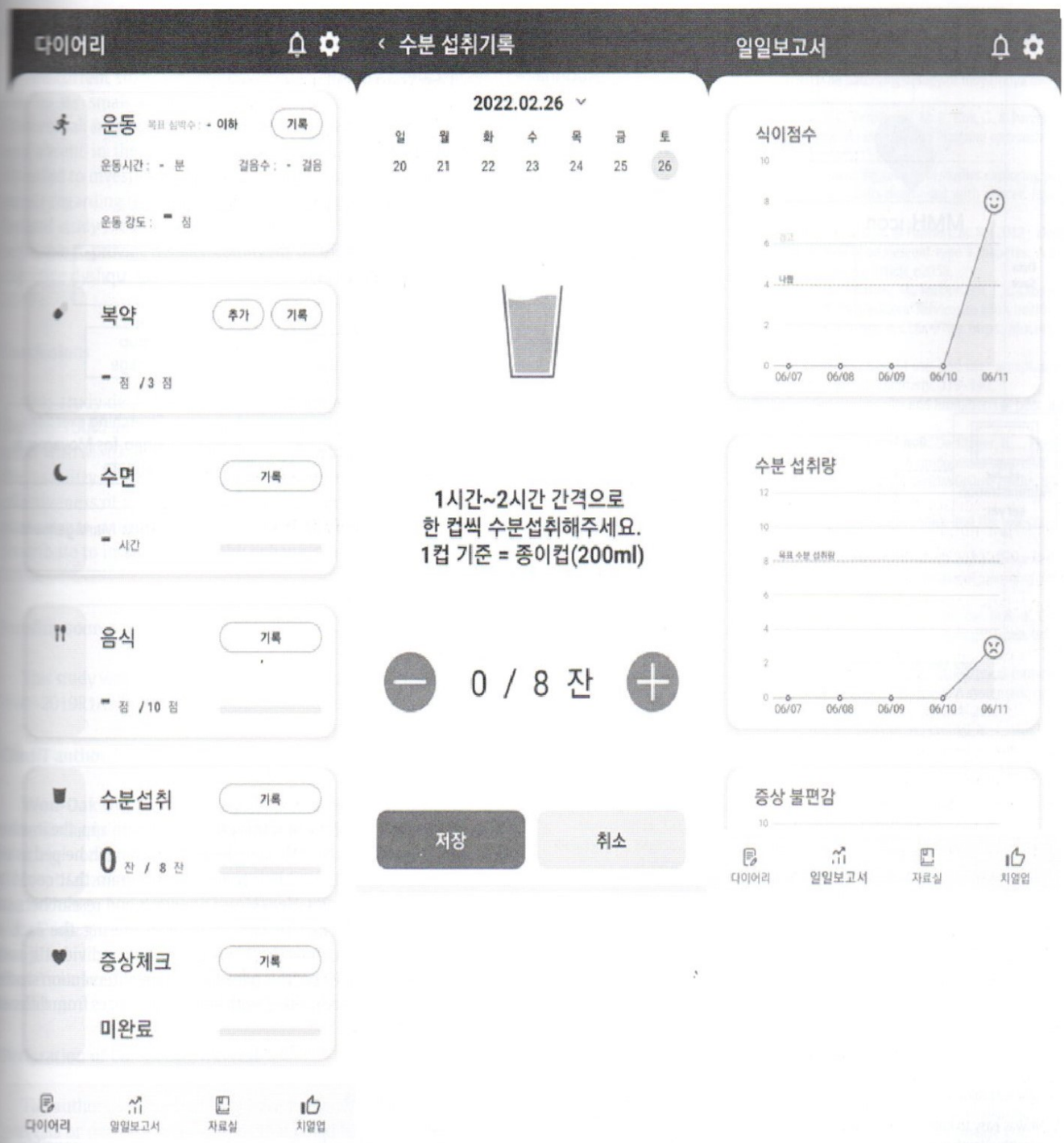

Fig. 2. Diary and diary report.

even a small amount of stress could trigger TIAs (Lee et al., 2017). Moreover, the app users demonstrated satisfaction with the actuation speed of the app. These results suggest that the Moyamoya Healthy Youth app is a promising method for enhancing the self-management skills of adolescents with MMD.

Despite the proliferation of smartphones as a healthcare device, to the best of our knowledge, this is the first attempt to systematically develop a mHealth program targeting adolescents with MMD, based on a robust theory and protocol. For adolescents with MMD, a close-at-hand device is indispensable to manage their symptoms since most of their daily activities immediately and critically influence cerebral blood flow, with potentially fatal results. In recent times, smartphone-use has led the current generation of adolescents to be called "digitally native". The Moyamoya Healthy Youth app program delivered by smartphones can thus play a pivotal role in the self-management of symptoms among adolescents with MMD. Especially, more vulnerable cases, who are less likely to get support from others can benefit from the Moyamoya Healthy Youth app, as it helps them manage their symptoms by themselves.

Furthermore, it should be noted that this app can be an appropriate and accessible way to promote the self-management behaviors of adolescents with MMD who are experiencing a transition from pediatric to adult healthcare systems. With this transition, they should begin taking responsibility for their own health behaviors, which is no longer their caregivers' responsibility. This transition period for adolescents with chronic illness, is often associated with worsening illness control and unplanned hospitalizations, attributable to inadequate self-management behaviors (Campbell et al., 2016). The Moyamoya Healthy Youth app is an invaluable resource to assist adolescents with MMD with navigating this transition.

Our work contributes to the field of adolescents with MMD and the development of an mHealth program for this population. As part of the

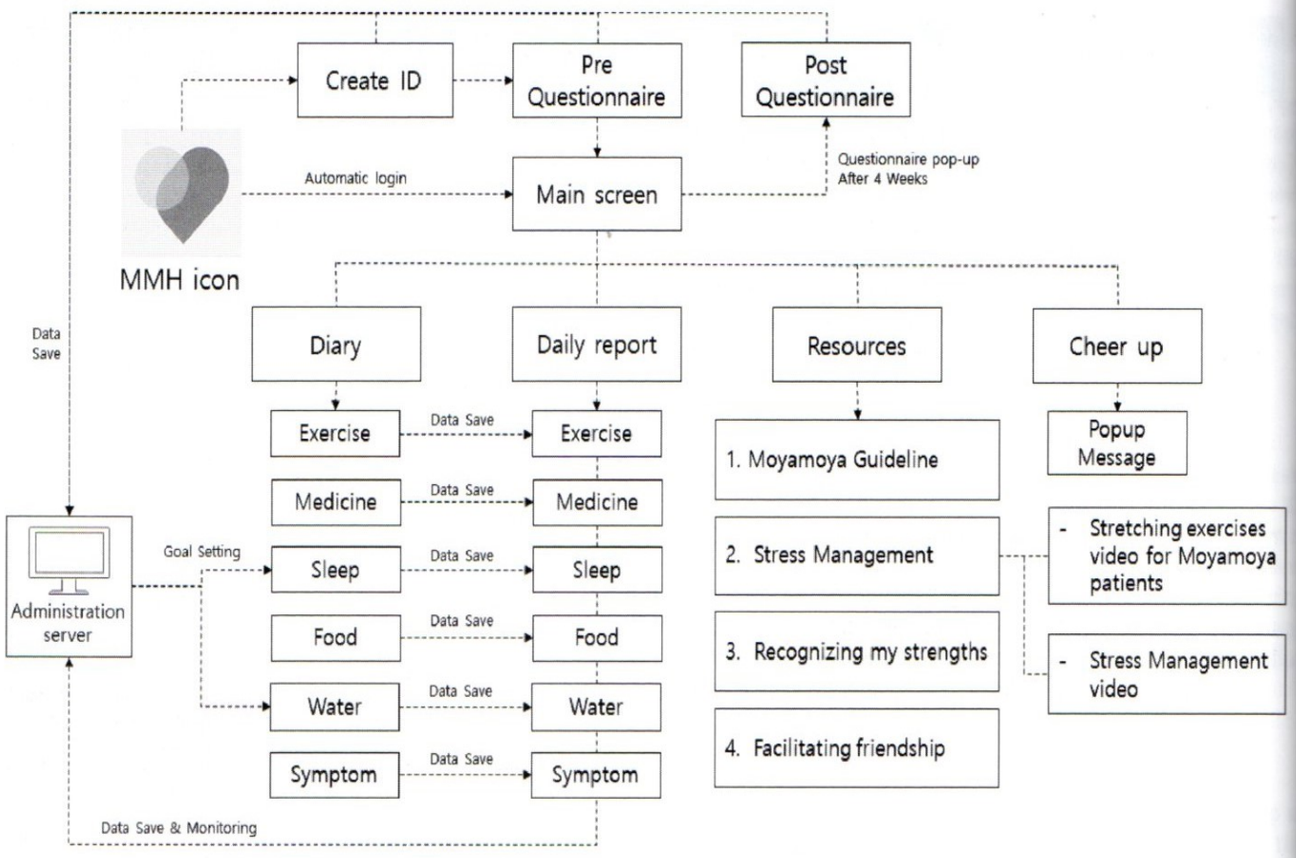

Fig. 3. Moyamoya Healthy Youth application Algorithm.

Table 2
Usability test of Moyamoya Heathy Youth application.

Items	Mean (SD)
Ease of use	39.75 (4.34)
It was easy to learn how to use the Moyamoya Healthy Youth app	4.50 (0.76)
The text information on the screen was clear	4.38 (0.92)
The text information on the screen was easy to read	4.38 (0.74)
It was easy to know what to do at each stage of the Moyamoya Healthy Youth app	4.38 (0.74)
I didn't have any technical problems using the Moyamoya Healthy Youth app	4.00 (0.93)
The visual quality of the video was good	4.25 (0.71)
It was fun using Moyamoya Healthy Youth app	4.00 (1.06)
The pace of the action was good	5.00 (0.00)
The audio quality of the video was good	4.88 (0.35)
Usefulness	27.13 (3.44)
I think that the Moyamoya Health Youth app will help adolescents with MMD improve the knowledge on MMD	4.63 (0.52)
I think that the Moyamoya Health Youth app will help adolescents with MMD improve their healthy behaviors	4.63 (0.52)
I think that the Moyamoya Health Youth app will help adolescents with MMD improve their quality of life	4.63 (0.52)
I think that the Moyamoya Health Youth app will help adolescents with MMD manage their stress	4.75 (0.46)
I think that the Moyamoya Health Youth app will help adolescents with MMD improve peer relationships	4.25 (0.89)
I think that the Moyamoya Health Youth app will help adolescents with MMD improve their self-esteem	4.25 (0.89)

development process of the Moyamoya Healthy Youth app, the involvement of adolescents with MMD and relevant professionals helped us develop an effective and feasible intervention program that could be replicated in future studies. Health care providers and researchers can apply the lessons learned in this study, considering the lack of evidence-based self-management programs for individuals with MMD. Based on the results of this pilot study, more intervention studies on the developed app are needed, with large sample sizes from different cultural backgrounds.

Practice implications

Nurses and other health care providers are responsible for empowering adolescents with MMD to enhance their self-management skills. The Moyamoya Healthy Youth app, developed based on a sound theoretical framework and the lived experiences of the target population, can be used as a promising method for this purpose. The usefulness and ease of use of the app were well-perceived by participants after using the app. Given that adolescents are digitally native, this method is expected to be highly applicable to adolescents with MMD.

Furthermore, the current project of the app development was supported by the Korean government as a part of the National Rare Disease project. Under the national initiative, there is great scope to use the app on a national scale to enable MMD patients in hospitals or communities to manage symptoms. If the Moyamoya Healthy Youth app becomes widely used, we plan to hire more app administrators to manage it. Although the app manager's monitoring is important to keep app users engaged with the app, our main intent is to enable app users to manage by themselves through the app's core functions of visualized data of their management status and pop-up messages. The Moyamoya Healthy Youth app can be a promising method for managing MMD symptoms at scale.

Limitations

The current study was not powered to examine treatment effects due to its small sample size, a common pitfall of pilot studies (Dworschak & Campbell, 2015). Additionally, the end-user involvement was absent in the pilot intervention study. In this pilot study, we intended to investigate the usability of the app and gain precise comments regarding the experience using it, before embarking on a full-fledged study for adolescents with MMD. Individuals with MMD may be unable to provide detailed comments on the app because of possible cognitive dysfunction due to cerebral hypoperfusion (Kazumata et al., 2019).

Conclusions

This study delineated the process of developing the Moyamoya Healthy Youth app to improve the self-management skills of adolescents with MMD. Additionally, the results of the pilot test confirmed the usability of the app. These results demonstrated the possible effectiveness of a self-management program using mHealth among adolescents with MMD. The Moyamoya Healthy Youth app can contribute to increasing healthy behaviors among adolescents with MMD.

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CRediT authorship contribution statement

Won-Oak Oh: Conceptualization, Funding acquisition, Investigation, Methodology, Project administration, Supervision, Validation, Writing – original draft, Writing – review & editing. **Il Tae Park:** Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Validation. **Jihee Han:** Conceptualization, Investigation, Project administration, Resources, Writing – original draft. **Eunji Lee:** Conceptualization, Data curation, Investigation, Validation. **Anna Lee:** Validation, Writing – original draft, Writing – review & editing.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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