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Phone-based motivational interviewing to increase self-efficacy in individuals with phenylketonuria

Krista S. Viau [a,b,*], Jessica L. Jones [b], Maureen A. Murtaugh [c], Lisa H. Gren [b], Joseph B. Stanford [b], Deborah A. Bilder [d]

[a] Department of Pediatrics, University of Utah, 295 Chipeta Way, Salt Lake City, UT 84108, USA
[b] Department of Family and Preventive Medicine, University of Utah, 375 Chipeta Way, Suite A, Salt Lake City, UT 84108, USA
[c] Department of Internal Medicine, University of Utah, 295 Chipeta Way, Salt Lake City, UT 84108, USA
[d] Department of Psychiatry, University of Utah, 650 Komas Drive, Suite 200, Salt Lake City, UT 84108, USA

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ABSTRACT

Objective: To measure change in patient activation and self-efficacy in individuals with phenylketonuria (PKU) before and after a 6-month phone-based motivational interviewing (MI) intervention and determine the feasibility of implementing dietary counseling for PKU using an MI approach.
Methods: Participants ($n = 31$) included preadolescents (7–12 years), adolescents (13–17 years), and adults (18–35 years) with early-treated PKU. Participants completed online questionnaires assessing self-reported stage of change (SOC), patient activation, and self-efficacy for PKU self-management behaviors. The intervention included monthly phone-based dietary counseling using MI during which participants set monthly goals.
Results: Patient activation and self-efficacy were significantly different by age group (both $p < 0.01$) with higher scores in older participants. Self-efficacy significantly increased from baseline to month 6 among adolescents and adults (7.4 ± 1.9 and 8.6 ± 1.3, respectively, $p = 0.002$). Preadolescents did not have a significant change in self-efficacy ($p = 0.79$). There was no increase in patient activation for preadolescents or adolescents/adults ($p = 0.19$ and $p = 0.24$, respectively). Indicators of learning problems were not significantly associated with self-efficacy ($p = 0.33$) or patient activation ($p = 0.83$).
Conclusion: These results demonstrate the feasibility of implementing phone-based dietary counseling for PKU using MI. This study also supports further investigation of MI as an intervention approach to improving self-efficacy and self-management behaviors in adolescents and adults with PKU.

1. Introduction

Poor treatment adherence is frequently reported among individuals experiencing chronic illness [1–4], including those with phenylketonuria (PKU). In individuals with PKU, there are multiple studies reporting rising blood phenylalanine (Phe) concentrations with age [5–7] and high rates of loss to follow up with only an estimated 23% of adults with PKU receiving treatment from a metabolic center in 2012 [8,9].

Adherence to daily medical and/or dietary regimens becomes increasingly more difficult as treatment complexity increases [10,11]. The ability to adhere to PKU treatment recommendations may be further impaired by subtle cognitive deficits or psychiatric symptoms [12,13]. Treatment of PKU is multifaceted, and successful management includes daily monitoring of dietary Phe or protein intake, consumption of a Phe-free medical formula, weekly to monthly monitoring of blood Phe levels, and regular food records and appointments with a metabolic clinic [14]. In a study of 19 families with PKU, all children reported difficulty following the diet and 91% reported problems drinking medical formula. Maladaptive strategies to address these difficulties were correlated with elevated blood Phe levels [15]. Strategies to facilitate behavior change are needed to improve treatment adherence in individuals with PKU.

Tailored interventions based on an individual's self-reported readiness to change have been shown to be more effective for improving health behavior change compared to more traditional approaches [16–20]. Stages of change (SOC), first described by James Prochaska and Carlo DiClemente [21], are a means of conceptualizing the process people experience as they evolve from having minimal insight and/or desire to change a behavior to the point at which they have changed this behavior and maintained this change. SOC reflect one's readiness to engage in behavior change [21,22]. Motivational interviewing (MI) is an intervention that incorporates a provider's understanding of their patient's current SOC to provide tailored counseling to support the patient's ability to progress towards the desired behavioral change. MI

Abbreviations: IEP, individualized education program; ID, intellectual disability; IQR, interquartile range; MI, motivational interviewing; PAM, Patient Activation Measure; Phe, phenylalanine; PKU, phenylketonuria; SOC, stages of change.

* Corresponding author at: 295 Chipeta Way, Salt Lake City, UT 84108, USA.
E-mail address: krista.viau@hsc.utah.edu (K.S. Viau).

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is a patient-centered, collaborative style of communication that elicits a patient's intrinsic motivation for change. MI employs strategies, such as reflective listening, supporting patient autonomy to decide whether or not to change, monitoring the patient's degree of readiness to change, affirming the patient's choices, and reducing resistance. The practitioner guides the patient and helps them identify discrepancies between their current actions and desired outcomes, creating opportunity for the patient to develop motivation to change [23–27].

In several chronic medical conditions, multiple studies and meta-analyses support the effectiveness of MI for improving self-efficacy, self-management behaviors, and health outcomes [28–33]. For example, one meta-analysis reported improved self-efficacy with MI in patients with diabetes, cardiovascular disease, or smoking, producing an overall effect size of 1.39 (95% CI 1.09-1.78) [31]. MI has been associated with subjective and objective measures of improved disease management including self-monitoring (blood sugar, food intake, and exercise), glycemic control, blood pressure, cholesterol, HIV viral load, and body weight [31,34,35]. MI has also demonstrated positive results in a variety of settings [28,36–38], and age groups [32,33,39,40].

Patient activation also appears to mediate health behavior change. Patient activation refers to the knowledge, skills, confidence, and motivation to manage one's own health [41]. Increased patient activation has been consistently associated with improved self-management behaviors, health outcomes, and reduced health care costs among individuals with a variety of chronic diseases [11,19,42–48]. Improvements in patient activation occur over a continuum and can be measured across self-care skills rather than linked to a single targeted behavior [49]. This characteristic lends itself to studying patient progress towards better disease self-management across multiple health behaviors related to this goal.

Like patient activation, increased self-efficacy coincides with improved health behaviors. Self-efficacy is the belief that one can successfully implement a behavior required to achieve the desired outcome [22]. Meta-analyses have reported higher self-efficacy is associated with increased participation in recommended self-care in adults with type 1 and 2 diabetes and adherence to antiretroviral therapy in individuals with HIV [11,48]. Self-efficacy appeared to be a critical component in moving from goal setting to goal completion in a study of adolescent females making healthy food choices [47]. Crone et al. found that increased parental self-efficacy regarding medical formula intake ($p = 0.007$) and raising a child with PKU ($p = 0.028$) was associated with improved blood Phe levels in their children [50]. Enhancing self-efficacy among individuals with PKU, as with other chronic diseases, may serve an important role in increasing treatment adherence related to PKU.

The aim of the current before-and-after study was to (1) measure change in patient activation and self-efficacy during the course of the intervention and (2) determine the feasibility of implementing dietary counseling for PKU using an MI approach. As a frontier state, Utah's population is spread over 85,000 mile^2. The catchment area of Utah's primary PKU clinic includes Utah, Idaho, and Wyoming. Because many patients residing in other states also face challenges related to the physical proximity of their closest PKU clinic, an additional component of this study was the implementation of intervention through phone contact, rather than in-person.

2. Methods

2.1. Participants

Study participants were individuals aged 7–35 years, diagnosed with PKU on newborn screening and treated within one month of birth, English speaking and had internet access at home. Exclusion criteria included 1) intellectual disability (IQ < 70), 2) pregnancy, as excess Phe is teratogenic and requires different treatment guidelines [51], 3) hyperphenylalaninemia not requiring dietary treatment, and 4) concurrent participation in clinical trial(s) testing enzyme substitution therapy. Patients were identified and recruited through the Utah Metabolic Clinic from December 2013 to July 2014. Participants were compensated monetarily for their time. The University of Utah Institutional Review Board approved this study and written consent – and child assent, if appropriate – was obtained for all participants.

2.2. Measures

2.2.1. Stages of change

A questionnaire based on a format previously shown to be reliable was compiled to assess participants' current SOC [52,53]. Treatment of PKU is complex and multiple behaviors contribute to blood Phe levels [54]. The following three behavioral domains were chosen to assess SOC: meeting dietary Phe/protein goals, reaching medical formula goals, and making healthy food choices. Although, healthy food choices may not affect Phe levels to the extent that Phe/protein and medical formula do, the inclusion of this domain provided opportunity to discuss other areas of improvement for those already meeting Phe/protein and formula goals. This goal also provided an option for participants not yet ready to discuss working towards Phe/protein or formula goals. Participants were asked to select the most important behavior from a list within each behavioral domain (Table 1). The participant could write in an additional behavior of interest if items listed were not felt to be pertinent. For each domain, the SOC was scored on a progressive scale from 1 (*absence of the desire to change behavior*, *precontemplation*) to 5 (*presence of the desire to maintain a changed behavior*, *maintenance*).

2.2.2. Patient activation

Activation was measured with the PAM-13. This questionnaire measures perceived knowledge, ability, and confidence to manage one's health [49]. PAM-13 scores range from 0 to 100, and this score can then be divided into four levels of activation [41]. These levels reflect a patient's belief that he/she should play an active role in self-care and collaborate with providers (level 1), knowledge about one's disease and its treatment (level 2), confidence to support new behaviors (level 3), and ability to maintain lifestyle changes in times of stress (level 4) [49]. The PAM-13 has been validated in adults, but not in children [49].

2.2.3. Self-efficacy

Self-efficacy was measured with a modified version of the 8-item Diabetes Self-Efficacy Scale developed at the Stanford Patient Education Research Center [55,56]. Items were ranked on a 10-point Likert scale ranging from 1 (*not confident*) to 10 (*totally confident*). The total score reflected the average of the eight items rather than the sum to maintain

Table 1
List of behavioral targets for stage of change questionnaire.

Domain	List of behaviors
Dietary Phe/protein	Count how much Phe/protein I eat
	Keep diet records
	Plan meals beforehand
	Watch my portion sizes
	Prepare meals at home
Medical formula	Drink all formula every day
	Drink formula several times per day
	Make my own formula
	Bring my formula to school/work
	Try a different formula or try to improve the taste of my current formula
Healthy food choices	Eat more fruits/vegetables
	Drink fewer sweetened drinks (soda, juice)
	Eat out less often (restaurants, fast food)
	Eat fewer "junk foods" (chips, cookies, candy)
	Cook meals at home more often

Phe, phenylalanine.
Note: Participants were asked to "Choose the single most important thing you could personally do to meet your goals" for each domain.