Benefits of Support Conferences for Parents of and People with Moebius Syndrome

Kathleen R. Bogart

Oregon State University

Amanda R. Hemmesch

St. Cloud State University

Author note

The first author of the study is a board member of the Moebius Syndrome Foundation, which may be perceived as a conflict of interest. However, the Moebius Syndrome Foundation is a non-profit organization, and the author receives no salary from the Foundation.

Correspondence concerning this article should be addressed to Kathleen Bogart, School of Psychological Science, Oregon State University, 2950 SW Jefferson Way, Corvallis, OR. Email: kathleen.bogart@oregonstate.edu

Published as: Bogart, K. R., & Hemmesch, A. R. (2016). Benefits of support conferences for parents of and people with Moebius syndrome. *Stigma and* *Health, 1*(2), 109-121. doi: 10.1037/sah0000018

**Abstract**

Hundreds of millions of people worldwide with rare diseases face unique challenges to quality of life (QoL), including stigma and limited support. To address these concerns, many rare disease organizations offer support conferences for people to meet others with their condition. This is the first research to examine the effects of a support conference for a rare disease, Moebius syndrome (MoS), a condition characterized by impaired facial and eye movement. Parents of and adults with MoS were predicted to receive different QoL benefits from support conferences. Individuals with MoS were predicted to receive *psychosocial benefits* including increased social comfort, emotional and companionship support, and reduced stigma, anxiety, and depression. Parents of people with MoS were predicted to gain *rare disease self-efficacy*, including increased informational and instrumental support, disability self-efficacy, or perceived competence in managing a disability, and knowledge about managing their child’s MoS. Forty-seven adults with MoS and 48 parents completed an internet-based survey four weeks prior to attending or not attending a support conference for MoS and six weeks after the conference. Correlational and pre-post analyses supported that adults with MoS received social comfort benefits, reduced stigma and increased perceived knowledge from support conferences, but no pre-post changes in anxiety, depression, or social support were detected. After applying a more conservative Bonferroni correction, only perceived knowledge remained significant. For parents, correlations suggested that repeated conference attendance was associated with rare disease self-efficacy and perceived knowledge, but no pre-post changes were detected. Rare disease support conferences are promising QoL interventions.

**Keywords**: rare diseases; stigma; social support; disability self-efficacy; Moebius syndrome

**Benefits of Support Conferences for Parents of and People with Moebius Syndrome**

People with rare diseases and disorders are underserved by health research due to a perception that efforts would impact only a small group of people. Although rare diseases differ in etiology and symptoms, many of the psychosocial experiences involved with having a rare disease are universal, including stigma and the need for specialized information and care. Researching these factors could ultimately help the estimated 350 million people worldwide who are living with one of 7,000 rare diseases (Walker, 2014).

Rare disorders create two dimensions of stigma which have not been addressed by current theory (Joachim & Acorn, 2003). First, rare diseases are isolating. Many people with rare diseases never have the normalizing opportunity to meet others like themselves (Limb, Nutt, & Sen, 2010). Second is the public’s lack of awareness. People with rare disorders are often burdened with the decision to explain their condition repeatedly or risk being misunderstood and not having their needs met (Bogart & Tickle-Degnen, 2015; Jaeger, Röjvik, & Berglund, 2015). The other unifying factor of rare diseases is that people with rare diseases seek information and healthcare through nontraditional pathways. General practitioners usually serve as gatekeepers for information and referrals, but people with rare diseases are most likely to get these resources from disease-specific patient organizations and support groups (Aymé, Kole, & Groft, 2008).

**Moebius Syndrome**

This research focuses on Moebius syndrome (MoS), a very rare congenital disorder, because it is the subject of a well-established support conference. MoS is characterized by facial paralysis and impaired lateral eye movement (Verzijl, van der Zwaag, Cruysberg, & Padberg, 2003). It can be traced to underdevelopment of the 6th and 7th cranial nerves, and occurs in 2 to 20 people per million (Verzijl et al., 2003). In most cases, it does not run in families (Verzijl et al., 2003), so parents typically learn about MoS for the first time when a child is diagnosed with it. A variety of other symptoms are sometimes present in those with MoS, such as limb or chest wall abnormalities and speech and eating difficulties.

MoS can be highly stigmatizing because people with MoS are frequently stereotyped as unfriendly, sad, or intellectually disabled (Bogart, 2014; Bogart & Tickle-Degnen, 2015; Bogart, Tickle-Degnen, & Ambady, 2014). MoS is visible but unrecognizable, meaning that strangers notice that the face looks unusual, but because most people have not heard of MoS, they cannot figure out why (Bogart & Tickle-Degnen, 2015). Strangers may become preoccupied with trying to figure out the cause or nature of the facial difference while also trying to avoid saying the wrong thing, which can result in awkward and ultimately stigmatizing interactions. Although day-to-day interactions can be stigmatizing, qualitative studies conducted at MoS conferences indicated that individuals with MoS value meeting others with their condition and feel socially comfortable in that setting (Bogart, Tickle-Degnen, & Joffe, 2012; Bogart, 2014).

**Social Support and Rare Diseases**

Approximately two-thirds of people with rare disease feel they do not get adequate social and psychological support (Limb et al., 2010). The focus of this paper was support quality, not quantity. While quantity can be an indicator of loneliness or isolation, social quality can provide more information about the processes, costs, and benefits of relationships, and has been associated with physical and psychological health (Cohen, 2004; Feeney & Collins, 2015). Social support can include different domains such as companionship support, like sharing pleasant activities; emotional support, like interactions with warmth, kindness, or encouragement; instrumental support, like assistance or aid for tangible tasks or goods; and informational support, like providing information and good advice (Walen & Lachman, 2000). In a large, representative sample of adults in the United States, these domains were associated with a variety of outcomes related to quality of life (QoL), including life satisfaction, mood, and health (Walen & Lachman, 2000). Individuals with stigmatizing conditions are more likely to seek a support group than those with less stigmatizing conditions (Davison, Pennebaker, & Dickerson, 2000), which suggests that individuals with rare diseases and their families may particularly crave social support.

The majority of research on social support interventions has focused on common medical conditions and the domain of emotional support, although the roles of instrumental and informational support have also been examined (Hogan, Linden, & Najarian, 2002). Support interventions have taken many forms, from relying on professional support (e.g., nurses) to recruiting existing support sources (e.g., family); and from using individual-based strategies to using group-based strategies (Hogan et al., 2002). Overall, support interventions seem somewhat effective in improving health and QoL outcomes, although different definitions, interventions, and populations are associated with differences in success (for a review, see Hogan et al., 2002). Research on peer-led group interventions, similar to some of the sessions provided during the MoS conferences, has shown that they can improve feelings of support and well-being (Hogan et al., 2002). In one of the only studies of a support intervention for the rare disease population, Dellve, Samuelsson, Tallborn, Fasth, and Hallberg (2006) found that an informational support program for parents about medical, social, and caregiving topics reduced parental distress over the course of a year.

Rare disease support conferences may be an ideal intervention for the unique challenges faced by this population, although they have not yet been studied. People with rare diseases and their families strongly desire meeting others with their condition (Huyard, 2009), but it is common to have never done so (Limb et al., 2010). There are usually too few resources and individuals in a particular geographic area to hold local support groups for a rare disease. Instead, many rare disease organizations organize annual or biannual weekend-long support conferences for people with rare conditions to meet others with their condition and experts familiar with their medical needs. People with rare diseases and their families often travel great distances to attend. The immersive nature of such conferences may be especially useful for reducing stigma and providing intensive informational opportunities. This type of intensive contact may help with feelings of isolation and foster lasting supportive relationships.

**Quality of Life Outcomes**

Our research aimed to explore many potential associations between conference attendance and QoL, a multi-faceted outcome that can include physical and psychological well-being, social support, stigma, and components that are unique to having a chronic or rare condition such as competence at managing a disease (Centers for Disease Control & Prevention, 2011; Peto, Jenkinson, Fitzpatrick, & Greenhall, 1995). We predicted that individuals with rare diseases and parents would receive different QoL benefits from support conferences, so we divided these components of QoL into two facets. Individuals with MoS were predicted to receive *psychosocial benefits* including increased social comfort, emotional and companionship support, as well as reduced stigma, anxiety, and depression from the normalizing experience of being surrounded by people with the same condition. On the other hand, we termed the potential benefits that parents of people with MoS were predicted to receive as *rare disease self-efficacy*, which included increased informational and instrumental support, disability self-efficacy, and knowledge about managing their child’s MoS.

There are several reasons to suggest adults and parents of people with rare disorders may experience different QoL benefits from support conferences. First, parents typically seek the initial knowledge, information and instrumental support for their child in the form of information about diagnosis, treatment, and services, with the goal of developing disability self-efficacy, which is defined as the feeling of competence in managing the tasks of the disability to achieve valued goals (Amtmann et al., 2012). Once the person with the condition has reached adulthood, it is likely that he or she has become an expert in managing the medical aspects of his or her symptoms, resulting in relatively stable rare disease self-efficacy. As an adult, psychosocial issues like companionship support, emotional support, anxiety, depression, and stigma may come to the forefront. Studies of psychological distress in MoS have found mixed results, with one finding increased anxiety and depression (Briegel, 2007), one finding no difference compared to the general public (Bogart & Matsumoto, 2010), and one finding less than the general population (Briegel, 2012). There are a number of reasons for inconsistent findings, including differences in measures, samples, and culture. A previously unexamined factor that may explain some of these discrepancies is connection to social support.

Second, people without disabilities tend to attribute the primary challenge of disability to the impairment, while people with disabilities are more likely to attribute the primary challenge to stigma and lack of accommodations (Olkin, 1999). Thus, parents may prioritize support for the physical symptoms, while adults may prioritize support for psychosocial issues. Supporting this notion, Huyard (2009) found that parents of children with rare diseases sought large advances in healthcare to cure their child, while adults with the condition sought improvements in QoL and modest improvements in healthcare.

**The Present Research**

This research focused on the Moebius Syndrome Foundation support conferences. The Moebius Syndrome Foundation is the only nonprofit organization in the US for MoS and is the largest in the world. It has been organizing biannual conferences since 1994 for a total of 11 conferences to date. The conferences last three days and rotate around the country. The most recent conference, which is discussed here, was held in July 2014 in Bethesda, Maryland. It provided opportunities for informational and instrumental support via structured talks by healthcare professionals and researchers about topics such as surgical intervention, genetics, employment, occupational therapy, speech therapy, ophthalmology, and psychology. Additionally, private consultations with expert healthcare providers were available. There were also many opportunities for emotional and companionship support. Several discussion groups were organized for specific populations, for example “just for moms,” “just for dads,” and “just for adults with MoS.” Meals were served at communal tables to be conducive to informal socializing. Additional social events were held at night. A total of 375 people attended the conference, including 122 parents of people with MoS and 44 adults with MoS (M. Abbott, personal communication, February 6, 2015). Also in attendance were other family members of people with MoS, healthcare professionals, and researchers.

We had two goals for this research. The first was to explore correlates of the total number of conferences attended. As there has been little research on this topic, these exploratory analyses could identify on the one hand, demographic factors associated with access to support conferences, and on the other hand, associations between repeated conference attendance and QoL. The second goal was to conduct a pre-post analysis of QoL outcomes among people who did and did not attend the most recent MoS conference. This analysis provided a more robust test of our prediction that adults with MoS would experience psychosocial benefits, while parents would receive rare disease self-efficacy from the conference.

**General Method**

**Overview**

This paper reports on two separate studies, adults with MoS (Study 1) and MoS parents (Study 2). These studies were conducted simultaneously using the same methodology and measures unless otherwise noted.

**Participants**

We sampled parents of people with MoS and adults with MoS separately, rather than parents and children from the same family. The rationale was that adults with MoS and parents could answer parallel measures, and adults with MoS would provide more accurate self-reports than children with MoS. Inclusion criteria were 18 years or older, could read and write in English, and self-reported having MoS or being a parent of someone with MoS. Participant characteristics are shown in Table 1.

**Procedure**

Oversight for the research was provided by the Oregon State University Institutional Review Board. Participants were recruited from the MoS Foundation mailing list and from MoS, facial paralysis, and rare disease support websites and social media. Recruitment materials indicated that researchers were interested in hearing from adults with MoS and parents of people with MoS who do and do not attend the MoS Foundation conferences. Potential participants were emailed a link 4 weeks prior to the conference. Potential participants were presented with an informed consent page on the survey administration website Qualtrics and indicated consent by clicking to proceed to the survey. Six weeks after the conference, participants were prompted to follow up via an email containing individualized Qualtrics links to the second survey. Six week follow-up has been used in other studies aimed to improve disease-related self-efficacy and was selected to increase retention (e.g., Tickle-Degnen, Ellis, Saint-Hilaire, Thomas, & Wagenaar, 2010). It took approximately 30 minutes to complete each survey.

**Measures**

Chronbach’s alphas for each measure are displayed in Tables 2 and 4.

**Social support.** Social support quality was measured with the 12-item Positive and Negative Social Exchange measure by Newsom, Rook, Nishishiba, Sorkin, and Mahan (2005). It was designed to measure four domains of social support based on work combining qualitative methods and confirmatory factor analysis (Newsom et al., 2005). Four subscales of social support were examined: companionship, emotional, informational and instrumental. The following are example items for companionship, emotional, informational, and instrumental support, respectively: “in the past month, how often did the people you know provide you with good company and support”; “cheer you up or help you feel better”; “offer helpful advice when you needed to make important decisions”; “provide you with aid and assistance.” Response options ranged on a 5-point scale from *never* to *very often*. Items were averaged, and higher numbers indicate greater support.

**Depression and anxiety.** The 14-item Hospital Anxiety and Depression Scale was used as a brief assessment of anxiety and depression (Zigmond & Snaith, 1983). It has been used with people with and without MoS and is unlikely to confound with physical symptoms (Bogart & Matsumoto, 2010). The following are example items for the depression and anxiety subscales, respectively, “I still enjoy the things I used to enjoy”; “I feel tense or ‘wound up.’” Possible scores for the anxiety and depression subscales range from 0 to 21, with higher numbers indicating greater anxiety and depression.

**Stigma.** Stigma was measured with the 21-item Perceived Stigma Questionnaire (Lawrence, Fauerbach, Heinberg, Doctor, & Thombs, 2006). This scale was developed for use among people with visible differences. It shows good convergent and discriminant validity with related constructs such as depression and social support (Lawrence et al., 2006). An example item is “People avoid looking at me.” Response options ranged on a 5-point scale from *never* to *always*. Items were averaged, with higher numbers indicating greater stigma. Parents were instructed to consider their child’s experience when answering the questions, as has been done by Lawrence et al. (2011).

**Disability self-efficacy.** Disability self-efficacy was measured with the University of Washington Disability Self-Efficacy Scale (Amtmann et al., 2012). This six-item Likert-type scale shows good psychometric properties and was designed for use with people with disabilities or chronic illness (Amtmann et al., 2012). The scale authors recommend replacing the word “disability” with a more specific condition when appropriate, so we used the term “Moebius syndrome.” There are no disability self-efficacy scales designed specifically for parents to our knowledge, so this scale was modified slightly when administered to our parent sample to refer to “your child’s Moebius syndrome” rather than “your Moebius syndrome.” Anexample item is: “you can [you can help your child] bounce back from frustration, discouragement or disappointment that Moebius may cause you [him/her]?” Response options ranged on a 5-point scale from *not at all* to *completely*. Higher numbers indicate greaterdisability self-efficacy. Possible scores range from 6-30.

**Perceived knowledge.** Perceived knowledge about MoS was measured with the following single item: “How knowledgeable do you feel about MoS?”

**Demographics.** Demographic variables including age, gender, income, education level, number of conferences attended, and whether they attended the most recent conference (at Time 2) were assessed.

**Research Design**

These studies were pre-post quasi-experiments. Participants self-selected to attend the conference in July 2014 or not.

**Data Analysis**

To determine whether there were differences between participants who remained in the study and those who left, *t* tests between these two groups were conducted for all demographic and outcome variables. Next, we conducted exploratory correlations using Pearson’s *r* between overall number of conferences attended at time 2, demographics, and the QoL variables described above. Pearson *r* also serves as an effect size measure for correlational analyses, with .10 denoting a small effect, .30 denoting a medium effect, and .50 denoting a large effect (J. Cohen, 1988). Next, pre-post analyses of outcomes for people who did and did not attend the 2014 MoS conference were conducted. ANCOVAs were calculated with the independent variable of conference attendance (attended this year or not)[[1]](#footnote-1) on each of the Time 2 dependent variables stigma, social comfort, emotional support, companionship support, informational support, instrumental support, depression, anxiety, knowledge, and disability self-efficacy, with the baseline measure of each dependent variable included as a covariate. As a conservative test to protect against familywise error, we also assessed the ANCOVA findings with a Bonferroni corrected alpha of .005. Cohen’s *d* effect sizes were reported for ANCOVAs. Values of .2, .5, and .8 correspond to small, medium, and large effects, respectively (J. Cohen, 1988).

**Study 1**

**Method**

Participants were adults with MoS (see Figure 1 for participant flow).In addition to the measures listed above, social comfort was measured with the 8-item Social Comfort Scale (Lawrence et al., 2006). This scale was developed for use among people with facial differences and shows good convergent and discriminant validity (Lawrence et al., 2006). An example item for the Social Comfort scale is “I feel like I fit in with most groups.” Response options ranged on a 5-point scale from *never* to *always*. Items were averaged, with higher numbers indicating greater social comfort scores. Only adults with MoS completed this scale because it has not been administered to parents regarding their children’s experience due to the subjective nature of social comfort.

**Results and Discussion**

Attrition analyses indicated no significant differences between participants who remained in the study and those who dropped out. Number of conferences attended was not significantly associated with any demographic variables. Providing some support for our hypothesis, correlations revealed that overall number of conferences attended was associated with a number of psychosocial variables including higher companionship support, emotional support, social comfort (marginally), and lower stigma (marginally; see Table 2). Overall conference attendance was associated with two other QoL variables that we did not predict: informational support and perceived knowledge.

ANCOVAs revealed improvements in stigma, *F*(1,43) = 6.32, *p* = .02, *d* = -.74, social comfort, *F*(1,43) = 6.43, *p* = .02, *d* = .76, and perceived knowledge, *F*(1,43) = 10.73, *p* < .01, *d* = .97 compared to people who did not attend. ANCOVAs did not approach significance for companionship support (*p* = .47, *d* = .21), emotional support (*p* = .20, *d* = .39), anxiety (*p* = .29, *d* = -.15), depression (*p* = .63, *d* = .32), informational support (*p* = .99, *d* = -.01), instrumental support (*p* = .93, *d* = .03), or disability self-efficacy (*p* = .40, *d* = .25). The only effect that remained significant with a more conservative Bonferroni corrected alpha was perceived knowledge. See Table 3 for *M* and *SD*. This provides some support for our prediction that psychosocial variables would improve after conference attendance compared to baseline.

**Study 2**

**Method**

Participants were parents of people with MoS (see Figure 2 for participant flow). In addition to the demographics listed above, participants reported the age and gender of their child.

**Results and Discussion**

Attrition analyses revealed that at baseline, individuals who later dropped out had less companionship support, *t*(55) = 2.94, *p* < .05, instrumental support, *t*(55) = 2.64, *p* = .01, and greater depression *t*(55) = -2.77, *p* < .01. Correlations (see Table 4) revealed that overall number of conferences attended was associated with several demographic variables, including greater age, child’s age (marginally), education, and income. Providing some support for our hypothesis, number of conferences attended was associated with disability self-efficacy and perceived knowledge. Additionally, attending more conferences was associated with two other QoL variables we did not predict: lower anxiety and depression (marginally).

None of the ANCOVAs examining the effects of attendance at the most recent conference on QoL approached significance (companionship support (p = .23, d = -.44) emotional support (p = .11, d = -.59) anxiety (p = .63, d = .17), depression (p = .39, d = -.32) stigma (p = .79, d = -.10), informational support (p = 85, d = -.07) instrumental support (p = .11, d = -.60) disability self-efficacy (p = .83, d = .08) and perceived knowledge (p = .86, d = -.06)). See Table 3 for *M* and *SD*. Thus, our hypothesis that parents would experience improved rare disease self-efficacy after the most recent conference was not supported.

**General Discussion**

This is the first research to our knowledge to examine the correlates and effects of attending a support conference for a rare disorder, MoS, on QoL. We expected that adults with MoS would receive psychosocial benefits, while parents would receive rare disease self-efficacy benefits.

**Psychosocial Outcomes**

In support of our prediction that conference attendance would be associated with psychosocial benefits for adults with MoS, repeated conference attendance was correlated with most of the psychosocial variables, including emotional support, companionship support, social comfort, and lower stigma. The predicted relationship between number of conferences attended and lower anxiety and depression was not supported for adults with MoS. Although we did not predict that repeated conference attendance would be associated with psychosocial outcomes for parents, it was associated with lower anxiety and marginally lower depression. This finding is consistent with previous research that has found that an informational program for parents of children with rare disorders reduced psychological distress (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006).

Turning to the pre-post outcomes of adults with MoS who did and did not attend the most recent conference, adults with MoS who attended the 2014 conference experienced improved social comfort and reduced stigma relative to baseline compared to adults with MoS who did not attend, supporting our hypothesis. None of the other hypothesized factors, companionship support, emotional support, anxiety, or depression, showed pre-post improvement in adults. These results offer converging evidence that some of the benefits of conference attendance can last at least six weeks. Additionally, the large effect sizes suggest that conference attendance had a substantial impact on psychosocial functioning for adults with MoS. People with rare disorders experience unique dimensions of stigma, including isolation and a lack of public awareness (Joachim & Acorn, 2003). Rare disease support conferences offer an opportunity where people with a condition can be surrounded by others who look like them and who understand their experiences. Support conferences also provide a weekend free from the burden of explaining one’s disorder to people who have never heard of it. The support conference model examined here is different from typical support interventions because it combines both peer- and professional-led support components. Furthermore, the weekend-long conference schedule is more intensive than many previously researched interventions that rely on shorter, more frequent contact (Hogan et al., 2002), which may make it better suited to the unique challenges of the rare disease community.

**Rare Disease Self-Efficacy Outcomes**

In contrast to our predictions for adults with MoS, we predicted that, for parents of people with MoS, conference attendance would be associated with rare disease self-efficacy. Half of the predicted correlations with conference attendance were supported in the parent data, including perceived knowledge and disability self-efficacy. The predicted relationship between number of conferences attended and informational and instrumental support was not found among parents.

Although we did not predict that conference attendance would be associated with rare disease self-efficacy for adults with MoS, the number of conferences adults with MoS attended was correlated with informational support and higher perceived knowledge. Adults with MoS who attended the most recent conference also experienced improved perceived knowledge compared to those who did not attend that year.

Although we predicted that parents would show improved disability self-efficacy, perceived knowledge, and informational and instrumental support after attending the 2014 conference, no pre-post change in QoL scores was observed after attending, relative to those who did not attend. There may be several reasons for this null result. First, the parent sample had a larger attrition rate than the adult MoS sample, and only 11 parents in the final sample did not attend the conference in 2014, meaning the study may have been underpowered. Second, opportunities for parents to gain rare disease self-efficacy may be pursued from rare disease organizations or online support groups, while the social benefits gained by adults with MoS may be unique to the normalizing conference experience. The potential for sustained rare disease self-efficacy from these resources may have weakened the ability to detect change after the 2014 conference.

**Demographic Factors Associated with Conference Attendance**

Another goal of this research was to explore the demographic factors associated with conference attendance, as this could identify groups who have less access to support conferences. While correlations revealed that demographic factors such as age, child’s age, income, and education were all associated with parental conference attendance, there were no significant relationships between demographic variables and conference attendance for adults with MoS. These factors may play less of a role for adults with MoS when considering whether to attend a support conference. Adults with MoS from a variety of demographic backgrounds may be more likely to attend than parents because the social benefits they receive are unique to the conference experience. On the other hand, parents with lower socioeconomic status may forego support conference attendance in favor of more easily accessible information regarding a rare condition, like through internet support sites. These results highlight the importance of outreach and funding efforts to help people in the rare disease community from a variety of backgrounds access support conferences.

**Limitations**

Sample size and representativeness are concerns in all rare disease research. It should be noted that these are the largest samples of adults with MoS and parents of children with MoS in a psychology study to date. Individuals not only self-selected membership in the organizations and participation in the study but also self-selected their recent conference attendance. As would be expected from a sample connected with rare disease organizations, our sample had a moderate to high socioeconomic status, had more women, and was largely white. Indeed, our findings may have been strengthened if our sample included a more representative socioeconomic range. People with low socioeconomic status may be at the most risk of QoL decrements, and thus may have the most potential for improvement. Because random assignment was not feasible for our design, a subject-expectancy effect is possible. That is, attendees’ self-reports at follow-up may have reflected pre-conceived expectations that they would benefit from the conference.

The parent study had more attrition than the study of adults with MoS, which may have left the final sample underpowered. Some of the parents who may have had the most to gain from the conference dropped out, those with lower companionship and instrumental support and higher depression. As such, we are less confident in our ability to detect effects of most recent conference attendance among parents than among adults with MoS. The parent study should be viewed as instructive for future research. Future studies may improve recruitment and retention by offering financial incentives for participation.

At the risk of increased Type 1 error, we took an exploratory approach by including a number of outcome variables in order to identify potential benefits of this understudied topic. We hope that the groundwork laid here will raise awareness of the need for funding and resources to conduct randomized trials of QoL interventions for people with rare diseases and their families using the outcomes identified in this study.

**Future Directions**

MoS shares universal characteristics with other rare diseases including stigma from isolation, a lack of public awareness, and specific informational needs. An important future direction would be to examine the role of support meetings for rare diseases with different courses and presentations, for example, ones that are invisible or progressive. People with invisible rare diseases may be more likely to attempt to “pass” by hiding their condition, making them less likely to seek support meetings. However, people with concealable stigma may have the most to gain from support meetings. They experience more psychological distress than those with visible stigma, and being around others who are known to share their stigma reduces this distress (Frable, Platt, & Hoey, 1998). Similarly, support conferences may be especially beneficial for people with progressive diseases. These individuals will contend with new and changing symptoms throughout their lives resulting in a dynamic striving for informational and instrumental support and disability self-efficacy.

**Implications**

These findings demonstrate the potential impact of rare disease support groups, particularly for individuals with those conditions. Repeated attendance, as well as attending the most recent conference, was associated with better QoL for individuals with MoS, and repeated attendance was associated with better rare disease self-efficacy for parents of individuals with MoS. The moderate-to-large effect sizes observed suggest that support conferences may offer useful opportunities for intervention for individuals with rare diseases and their families, although randomized controlled trials will be necessary to confirm these conclusions. Conferences may be ideal for improving social comfort and stigma because of the extended period of immersion. However, any sort of support meeting, including casual, patient-organized get-togethers or retreats for a combination of rare disorders may have beneficial social effects for people with rare diseases and improve rare disease self-efficacy for parents of people with rare diseases. Clinicians should utilize umbrella organizations for rare diseases such as the National Organization for Rare Diseases in the US and EURORDIS in Europe as referral networks to connect patients and their families with support meetings. More funding and resources are needed to research interventions to support the QoL needs of people with rare diseases and their families and to enable patients and their families to access these services.

**References**

Amtmann, D., Bamer, A. M., Cook, K. F., Askew, R. L., Noonan, V. K., & Brockway, J. A. (2012). University of Washington self-efficacy scale: A new self-efficacy scale for people with disabilities. *Archives of Physical Medicine and Rehabilitation*, *93*(10), 1757–1765. doi:10.1016/j.apmr.2012.05.001

Aymé, S., Kole, A., & Groft, S. (2008). Empowerment of patients: lessons from the rare diseases community. *The Lancet*, *371*(9629), 2048–2051. doi:10.1016/S0140-6736(08)60875-2

Bogart, K. R. (2014). “People are all about appearances”: A focus group of teenagers with Moebius Syndrome. *Journal of Health Psychology*, Advance online publication. doi:10.1177/1359105313517277

Bogart, K. R., & Matsumoto, D. (2010). Living with Moebius syndrome: Adjustment, social competence, and satisfaction with life. *The Cleft Palate-Craniofacial Journal*, *47*(2), 134–142. doi:10.1597/08-257.1

Bogart, K. R., & Tickle-Degnen, L. (2015). Looking beyond the face: A training to improve perceivers’ impressions of people with facial paralysis. *Patient Education and Counseling*, *98*(2), 251–256. doi:10.1016/j.pec.2014.09.010

Bogart, K. R., Tickle-Degnen, L., & Ambady, N. (2014). Communicating Without the Face: Holistic Perception of Emotions of People With Facial Paralysis. *Basic and Applied Social Psychology*, *36*(4), 309–320. doi:10.1080/01973533.2014.917973

Bogart, K. R., Tickle-Degnen, L., & Joffe, M. S. (2012). Social interaction experiences of adults with Moebius Syndrome: A focus group. *Journal of Health Psychology*, *17*(8), 1212–1222. doi:10.1177/1359105311432491

Briegel, W. (2007). Psychopathology and personality aspects of adults with Möbius sequence. *Clinical Genetics*, *71*(4), 376–377. doi:10.1111/j.1399-0004.2007.00787.x

Briegel, W. (2012). Self-perception of children and adolescents with Möbius sequence. *Research in Developmental Disabilities*, *33*(1), 54–59. doi:10.1016/j.ridd.2011.08.013

Centers for Disease Control - Concept - Health-Related Quality of Life. (n.d.). Retrieved June 22, 2015, from http://www.cdc.gov/hrqol/concept.htm

Cohen, J. (1988). Statistical power for the social sciences. *Hillsdale, NJ: Laurence Erlbaum and Associates*.

Cohen, S. (2004). Social relationships and health. *American Psychologist*, *59*(8), 676-684. doi:10.1037/0003-066X.59.8.676

Davison, K. P., Pennebaker, J. W., & Dickerson, S. S. (2000). Who talks? The social psychology of illness support groups. *American Psychologist*, *55*(2), 205-217. doi: 10.1037/0003-066X.55.2.205

Dellve, L., Samuelsson, L., Tallborn, A., Fasth, A., & Hallberg, L. R.-M. (2006). Stress and well-being among parents of children with rare diseases: a prospective intervention study. *Journal of Advanced Nursing*, *53*(4), 392–402. doi:10.1111/j.1365-2648.2006.03736.x

Feeney, B. C., & Collins, N. L. (2015). A New Look at Social Support A Theoretical Perspective on Thriving Through Relationships. *Personality and Social Psychology Review*, *19*(2), 113–147. doi:10.1177/1088868314544222

Frable, D. E., Platt, L., & Hoey, S. (1998). Concealable stigmas and positive self-perceptions: feeling better around similar others. *Journal of Personality and Social Psychology*, *74*(4), 909-922. doi:10.1037/0022-3514.74.4.909

Hogan, B. E., Linden, W., & Najarian, B. (2002). Social support interventions: Do they work? *Clinical Psychology Review*, *22*(3), 381–440.

Huyard, C. (2009). What, if anything, is specific about having a rare disorder? Patients’ judgements on being ill and being rare. *Health Expectations*, *12*(4), 361–370. doi:10.1111/j.1369-7625.2009.00552.x

Jaeger, G., Röjvik, A., & Berglund, B. (2015). Participation in society for people with a rare diagnosis. *Disability and Health Journal*, *8*(1), 44–50. doi:10.1016/j.dhjo.2014.07.004

Joachim, G., & Acorn, S. (2003). Life with a rare chronic disease: the scleroderma experience. *Journal of Advanced Nursing*, *42*(6), 598–606. doi:10.1046/j.1365-2648.2003.02663.x

Lawrence, J. W., Fauerbach, J. A., Heinberg, L. J., Doctor, M., & Thombs, B. D. (2006). The reliability and validity of the Perceived Stigmatization Questionnaire (PSQ) and the Social Comfort Questionnaire (SCQ) among an adult burn survivor sample. *Psychological Assessment*, *18*(1), 106. doi:10.1037/1040-3590.18.1.106

Limb, L., Nutt, S., & Sen, A. (2010). Experiences of Rare Diseases: An Insight from patients and families. Rare Disease UK. Retrieved from http://www.raredisease.org.uk/documents/RDUK-Family-Report.pdf

Newsom, J. T., Rook, K. S., Nishishiba, M., Sorkin, D. H., & Mahan, T. L. (2005). Understanding the relative importance of positive and negative social exchanges: Examining specific domains and appraisals. *The Journals of Gerontology Series B: Psychological Sciences and Social Sciences*, *60*(6), 304–312. doi:10.1093/geronb/60.6.P304

Olkin, R. (1999). *What psychotherapists should know about disability*. New York, NY: The Guilford Press.

Peto, V., Jenkinson, C., Fitzpatrick, R., & Greenhall, R. (1995). The development and validation of a short measure of functioning and well being for individuals with Parkinson’s disease. *Quality of Life Research*, *4*(3), 241–248.

Tickle-Degnen, L., Ellis, T., Saint-Hilaire, M. H., Thomas, C. A., & Wagenaar, R. C. (2010). Self-management rehabilitation and health-related quality of life in Parkinson’s disease: A randomized controlled trial. *Movement Disorders*, *25*(2), 194–204.

Verzijl, H. T. F. ., van der Zwaag, B., Cruysberg, J. R. ., & Padberg, G. W. (2003). M\öbius syndrome redefined. *Neurology*, *61*(3), 327-333.

Walen, H. R., & Lachman, M. E. (2000). Social support and strain from partner, family, and friends: Costs and benefits for men and women in adulthood. *Journal of Social and Personal Relationships*, *17*(1), 5–30.

Walker, K. K. (2014). Cognitive and affective uses of a thoracic outlet syndrome Facebook support group. *Health Communication*, *29*(8), 773–781. doi:10.1080/10410236.2013.800830

Zigmond, A. S., & Snaith, R. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica*, *67*(6), 361–370. doi:10.1111/j.1600-0447.1983.tb09716.x

Table 1

*Participant Characteristics*

**

Table 2

*Intercorrelations of demographic, psychosocial, and rare disease self-efficacy variables for adults with Moebius syndrome*



*Note*. \*\* *p* < .01, \* *p* < .05, † *p* < .10

Table 3

*Baseline and follow-up descriptive statistics for quality of life variables among parents of and adults with Moebius syndrome*

**

Table 4

*Intercorrelations of demographic, psychosocial, and information self-efficacy variables for parents of people with Moebius syndrome*



*Note*. \*\* *p* < .01, \* *p* < .05, † *p* < .10

1. Although it would have been interesting to compare participants who attended the conference for the first time in 2014 to those who attended in the past and those who never attended in the MANCOVA, we were not able to do so because it would create very small cells. For example, there were only three MoS participants who attended the conference for the first time in 2014. However, the correlational analyses involving the total number of conferences attended provide some insight into cumulative effects of the conference. [↑](#footnote-ref-1)