A Biopsychosocial Intervention for Parents and Caregivers of Children with Neurofibromatosis to Increase Self-Efficacy: A Pilot Study

-

Graduate School of Social Work and Social Research, Bryn Mawr College

SOWK B503: Research Informed Practice I

Professor Lauren Montemuro-Rode

December 18, 2020

Specific Aims

The specific aim of this pilot study is to provide a biopsychosocial intervention to parents and caregivers of children with Neurofibromatosis 1 (NF1) in order to increase their self-efficacy. The independent variable will be the intervention and the dependent variable will be parental and caregiver self-efficacy. Participants in this study will be parents or caregivers of children who seek medical treatment for NF1 at the Children's Hospital of Philadelphia. NF1 is a genetic condition that can significantly impact an individual's mental and physical health, education and psychological adjustment in a multitude of ways, throughout their entire life span (CTF, 2020). Qualitative research reveals that the variable nature of the progression of the condition, along with the potential to significantly impact one's appearance due to tumor growth, can negatively affect the quality of life and psychological adjustment of both child and caregiver (Rietman et al., 2018). Parents and caregivers of children with NF1 have high levels of stress and worry, difficulties knowing their parental role and concerns about the future for their child (Rietman et al., 2018). The qualitative research demonstrates a serious need to provide support and assistance to those affected by NF1. Currently, there is little to no quantitative research to both address the needs for those caring for children with NF1 and to increase knowledge, self-efficacy and ultimately the well-being of those affected by NF1. The biopsychosocial intervention will be given over an 8-week period, once a week in the evenings, and will seek to increase parents' and caregivers' self-efficacy, medical knowledge, and coping skills when working and caring for children with NF1.

Background

NF1, is a genetic and neurological disorder that affects 1 in every 3,000 children born worldwide (CTF, 2020). NF1 can be inherited from a parent who has the disorder, or in approximately half the cases, a spontaneous mutation-- a random error in the process of copying genetic information (CTF, 2020).

NF1 is often detected by multiple, more than six, café au lait skin spots on the body (Barke, et al., 2016). NF1 can present itself differently, from mild cases to more severe, which can change throughout the lifespan, from patient to patient (NINDS, 2020). Children with NF1 have many physical features and symptoms, such as softening of the bones and spine (scoliosis), optic gliomas, a large head circumference, small stature, high blood pressure and dermal neurofibromas, on the skin or under the skin (CTF, 2020). Tumors known as neurofibromas can grow on nerve endings and develop on or under the skin (NINDS, 2020). These neurofibromas in about 10% of cases can become malignant (CTF, 2020).

Up to 65% of children with NF1 have learning difficulties to varying degrees (Cutting et al., 2004). Additionally, many children with NFI experience attention deficit hyperactivity disorder, autistic spectrum disorders, lowered social skills and difficulties processing social information (Lehtonen et al., 2013).

A qualitative research study was conducted in the Netherlands by Rietman and colleagues (2018) exploring the worries and care needs in medical, psychological and socioeconomic domains of adults and parents of adults with NF1. Group interviews and focus groups were conducted which included patients (*n*=30) aged 18-67 years and parents of patients (*n*=12). Convenience sampling was used and research was done after the Neurofibromatosis Patient Organization of The Netherlands (NFVN) conference. This meeting is usually attended by between 50 to 100 patients, parents of patients and professionals. Findings from the research were significant. Parents reported high stress levels of anxiety, difficulties knowing their parental role, and fears about the future (Rietman et al., 2018). Difficulties with acceptance and coping with the disease was also widely present and various patients stated that it would have been helpful if psychological care had been offered throughout their NF1 journey (Rietman et al., 2018). There were various limitations to this study. All participants were members of the NFVN allowing a possible bias towards more well-informed patients and parents. Patients who participated may also experience more severe observable symptoms compared to other NF1 cases. Sample size was also limited. (Rietman et al., 2018). Rietman and colleagues reported an open and stimulating environment though participants may have felt uncomfortable expressing intimate matters, or deep feelings or concerns in the presence of other participants (Rietman et al., 2018).

Barke, Coad and Harcourt (2016) identified a gap in understanding specifically parents' and caregivers' experiences of caring for a child with NF1 and in their qualitative study, they explored these experiences. Convenience sampling was used and the seven parents that participated in the study were either from social media, online forums or recruited from the Neuro Foundation Newsletter (a UK-based support group for people with NF1 and their families) and they were interviewed by phone or in person. All parents had children ages 14 and above. Three key themes were identified after data was collected: "uncertainty and diversity", "appearance and noticeability" and "information and understanding" (Barke et al., 2016). Parents explained that the diagnosis caused great uncertainty and concern about the future. (Barke et al., 2016). Parents also reported appearance changes to their child with NF1 and managing others' comments, were also challenges they faced (Barke et al., 2016). Additionally, parents indicated that managing the unpredictability of NF1 and the diverse ways it can impact a child were key concerns for parents (Barke et al., 2016).

Both these qualitative studies explored parents' and caregivers' experiences of caring for a child with NF1 and have identified a strong need to further provide assistance and support for those effected by NF1. Minimal quantitative research for this population has been conducted. Also, the recent qualitative research that has been done has only focused on parents or caregivers of children 14 years of age and older. However, 85% of NF1 cases are diagnosed by age 6, and 95% are diagnosed by age 8 (BCH, 2020).

This quantitative research study looks to close this gap in research and positively impact the NF1 community. The biopsychosocial intervention will be available to parents and caregivers of children ages 4 years and older and provide information, intervention and support for those effected by NF1.

Conceptual Framework

The theory informing this research is based on Albert Bandura's 1977 theory of self-efficacy. Self-efficacy is a belief in one's ability to successfully perform a given task. Self-efficacy can help inform how an individual may behave, how much effort they put into a task and how long they persist in the face of adversity (Wittkowsi et al., 2017). Derived from Bandura's self-efficacy theory, research has looked specifically at parenting self-efficacy (PSE). PSE can be defined as a caregivers' or parents' confidence in their ability to successfully raise children (Coleman and Karracker, 1998). Higher PSE is strongly correlated with positive parent and child psychological functioning, child adjustment, parenting competence and parenting satisfaction (Jones and Prinz, 2005). There is also evidence that higher levels of PSE are strongly associated with "an adaptive, stimulating and nurturing child-rearing environment, which encourages social, academic and psychological well-being" (Wittkowski et al., 2017). Group-based parenting intervention programs have shown to positively influence PSE (Wittkowski et al., 2016). Based on my conceptual framework, I expect the biopsychosocial intervention will increase parents PSE and support a more positive, nurturing environment for parents, caregivers and children affected by NF1.

Methods

Measurements

The dependent variable, self-efficacy will be measured using the Parenting Empowerment and Efficacy Measure (PEEM), (Freiberg et al., 2014). PEEM is a strengths-based measurement tool and its main focus is on parents' and caregivers' sense of control or capability to confidently handle the challenges of being a parent or caregiver. PEEM invites parents and caregivers to identify their strengths and any influences in the environment or community that may help or hinder their capacity to successfully meet their parenting goals. (Freiberg et al., 2014). PEEM consists of 20 positively worded statements. Respondents are asked to use a 10-point scale to indicate how well each statement captures the way they feel about themselves as a parent. A rating of 1 indicates that the statement does not represent how they feel about themselves as a parent. A rating of 10 on the other hand, indicates that the statement is an excellent match and describes how they feel. No items are reversed scored and scores can range from 20 to 200. Higher scores represent greater amounts of parenting empowerment and efficacy (Freiberg et al., 2014). Analysis of the PEEM revealed high internal consistency ($\alpha = 0.92$) that correlated at 0.60 or more with three validation measures, as well as the existence of two hypothesized subfactors (correlated at 0.78): efficacy to parent and efficacy to connect, each with internal consistencies of 0.85+. Test–retest reliability (n= 200) was 0.84. PEEM exhibits excellent convergent and concurrent validity and is a reliable tool for use in planning services, monitoring participant progress, and evaluating program effectiveness. Social desirability was also assessed on the correlation between the PEEM score and the Marlow-Crowne Social Desirability Scale (MCSDS) (Freiberg et al, 2014). The correlation between the measures was low (*r*=-0.27; p=0.00; n =474) and comparable to the correlations between the social desirability measure and the validation measures. PEEM is not particularly susceptible to social desirability (Freiberg et al 2014).

Sampling and Recruitment

Participants in this study will be parents and caregivers of children ages 4-17 with NF1. Children must have a confirmed DNA diagnosis and seek treatment at Children's Hospital of Philadelphia. Parents or caregivers who have NF1 are not excluded from the study. Caregivers who are paid will be excluded from the study and a maximum of three parents or caregivers can be represented per child.

Prior to selection, the research study will be evaluated by CHOP's Institutional Review Board and Bryn Mawr College. Convenience sampling will be used in close cooperation with CHOP's Neurofibromatosis Pediatric Program. Social Worker and members of the NF1 pediatric team will obtain HIPAA and PHI consent forms to allow researchers to contact potential participants through email and phone, and to ensure confidentiality. A flyer will be placed at CHOP in the NF1 clinic to give details of the study: intervention, purpose of the study, time commitment required along with contact information. Participants will be invited to engage in an 8-week long biopsychosocial intervention. The Parent Empowerment and Efficacy (PEEM) survey will be given out four different times throughout the study and participants will independently fill out the survey and confidentiality in data collected will be maintained throughout the study.

The sample size for this study will be 15 to 20 participants. Biopsychosocial intervention will be 120 minutes, once per week, in the evenings, over an 8-week period. Intervention will take place at CHOP at the King of Prussia location. Participants will receive the PEEM survey as a pre-test, prior to the intervention. This data will be collected on the first day of the intervention and participants will be asked to come 20 minutes prior to class. PEEM survey data will be collected an additional two times, the beginning of the fourth class, and the last class. An additional PEEM will be administered via email after three months post class. Data will be evaluated to understand if there is an increase in self-efficacy post intervention.

Intervention will be led weekly and by the same two social workers who collaborated in creating the biopsychosocial intervention. Two physicians on the NF1 pediatric program team will also contribute in creating curriculum. Free child care will be provided by CHOP for children, and dinners of pizza and sandwiches will be provided by CHOP. The intervention will seek to increase self-efficacy, medical knowledge and coping skills among participants. Participants will receive a $10 gift card after each survey is taken.

Conclusion

NF1 is an unpredictable genetic condition that can cause a wide range of physical and mental health symptoms throughout one's life. It can cause both patients and family members significant anxiety, worry, stress and fear (Rietman et al., 2018). Parents and caregivers have also reported questioning their parenting ability and how to confidently provide support and information to their children regarding NF1 (Barke et al., 2016). Quantitative research is lacking, and minimal interventions have been provided for parent's navigating this condition. My pilot study looks to provide a biopsychosocial intervention to give medical information, coping skills, support and self-efficacy to parents and caregivers of children with NFI.

References

Barke, J., Coad, J., & Harcourt, D. (2016). Parents' experiences of caring for a young person with neurofibromatosis type 1 (NF1): a qualitative study. *Journal of community genetics*, *7*(1), 33–39. https://doi.org/10.1007/s12687-015-0247-z

Bandura, A. (1977). Self-efficacy: toward a unifying theory of behavioral change. *Psychological Review, 84*(2), 191-215. doi:10.1037/0033-295x.84.2.191

Children's Tumor Foundation. (2020). retrieved from https://www.ctf.org/

Neurofibromatosis: symptoms & causes: Boston Children's Hospital. (2020). Retrieved from https://www.childrenshospital.org/conditions-and-treatments/conditions/n/neurofibromatosis/symptoms-and-causes

Coleman, P. K., & Karraker, K. H. (1998). Self-efficacy and parenting quality: findings and Future Applications. *Developmental review, 18*(1), 47-85. doi:10.1006/drev.1997.0448

Cutting, L. E., Clements, A. M., Lightman, A. D., Yerby-Hammack, P. D., & Denckla, M. B. (2004). Cognitive profile of neurofibromatosis type 1: rethinking nonverbal learning disabilities. *Learning Disabilities Research and Practice, 19*(3), 155-165. doi:10.1111/j.1540-5826.2004.00099.x

Freiberg, K., Homel, R., & Branch, S. (2014). The parent empowerment and efficacy measure (PEEM): a tool for strengthening the accountability and effectiveness of family support services. *Australian Social Work, 67*(3), 405-418. doi:10.1080/0312407x.2014.902980

Jones, T. L., & Prinz, R. J. (2005). Potential roles of parental self-efficacy in parent and child adjustment: a review. *Clinical Psychology Review, 25*(3), 341-363. doi:10.1016/j.cpr.2004.12.004

Lehtonen A, Howie E, Trump D, Huson SM. Behaviour in children with neurofibromatosis type 1: cognition, executive function, attention, emotion, and social competence. Dev Med Child Neurol. 2013 Feb;55(2):111-25. doi: 10.1111/j.1469-8749.2012.04399.x. Epub 2012 Aug 31. PMID: 22934576.

Mouridsen, S. E., & Sorensen, S. A. (1995). Psychological aspects of von Recklinghausen neurofibromatosis (NF1). *Journal of Medical Genetics, 32*(12), 921-924. doi:10.1136/jmg.32.12.921

Neurofibromatosis Fact Sheet. (2020). Retrieved from https://www.ninds.nih.gov/disorders/patient-caregiver-education/fact-sheets/neurofibromatosis-fact-sheet

Rietman, A. B., Helden, H. V., Both, P. H., Taal, W., Legerstee, J. S., Staa, A. V., . . . Oostenbrink, R. (2018). Worries and needs of adults and parents of adults with neurofibromatosis type 1. *American Journal of Medical Genetics Part A, 176*(5), 1150-1160. doi:10.1002/ajmg.a.38680

Washington University Neurofibromatosis Center. (2020, November 13). Retrieved November 14, 2020, from https://nfcenter.wustl.edu/

Wittkowski, A., Dowling, H., & Smith, D. M. (2016). Does Engaging in a Group-Based Intervention Increase Parental Self-efficacy in Parents of Preschool Children? A Systematic Review of the Current Literature. *Journal of Child and Family Studies, 25*(11), 3173-3191. doi:10.1007/s10826-016-0464-z

Wittkowski, A., Garrett, C., Calam, R., & Weisberg, D. (2017). Self-Report Measures of Parental Self-Efficacy: A Systematic Review of the Current Literature. *Journal of Child and Family Studies, 26*(11), 2960-2978. doi:10.1007/s10826-017-0830-5