

**Understanding Barriers to Using Noninvasive Ventilation Through the Experiences of
Caregivers of Pediatric Patients with Neuromuscular Disorders**

Regan Bergan

College of Nursing, University of Tennessee - Knoxville

N457 Honors Nursing Leadership

Laurie Acred-Natelson, MSN, RN, NEA-BC

April 16, 2025

Understanding the Experiences of Caregivers of Pediatric Patients with Neuromuscular Disorders Using Noninvasive Ventilation

My clinical partner has over 25 years of experience in nursing with a focus on pediatric respiratory health. In her numerous years of experience, she noted a lack of adherence with treatment regimens regarding outpatient noninvasive ventilation (NIV). Without oxygen support, these children had to progress to more invasive ventilatory forms. She, along with her fellow clinicians, began to explore the deeper reasoning behind the nonadherence they were seeing. The objective of the following research is to understand barriers to the use of NIV as respiratory support for pediatric patients with a neuromuscular disorder (NMD) in an outpatient setting through the experiences of their caregivers.

PICO: How do caregivers of pediatric patients diagnosed with a neuromuscular disorder perceive barriers to the use of noninvasive ventilation as respiratory support?

Children with NMDs have weakened muscle tone throughout their bodies including the muscles of the respiratory system. As a result of this, they often experience poor gas exchange and perfusion. Rather than jumping to aggressive and invasive measures, physicians use NIV to improve the respiratory status of these patients (AlBalawi, et al., 2022; Fauroux, et al., 2020). By using the various forms of NIV, the flow of oxygen can be adjusted as needed for each client (Diaz-Macinnis, 2020). These devices affect inspiration and expiration depending on the pressure of the flow; they come in the form of a nasal cannula or masks with a wide range of sizes covering the nose, the mouth, the nose and mouth, or the whole face. These children are required to wear these devices either at night or all of the time. The NMDs frequently discussed

in the literature related to NIV are Duchenne Muscular Dystrophy (DMD) (Pascoe, et al., 2019) and Spinal Muscular Atrophy (SMA) (Al-Subu, et al., 2023; Schroth, 2009).

Previous studies have noted complaints related to the nature of the masks. Positive airway pressure devices are the most common NIV used in children with NMDs and therefore the devices most often seen in the research. These devices administer oxygen through masks that cover varying amounts of the face. Krivec and Caggiano (2020) report that one of the barriers to adherence with respiratory regimens was participant claims of claustrophobia. To deliver oxygen effectively, the devices must be covering some portions of the nose or mouth or both which may lead to feelings of smothering or entrapment. Where these devices are secured to the face, head, or around the ears, skin breakdown often occurs (Cammarota, et al., 2022; Fauroux, et al., 2020; Knebel, et al., 1997). The devices must be tight enough to prevent air leakage around the mask. However, this tightness often leads to continuous pressure in certain areas such as behind the ears and at the bridge of the nose. Such pressure can lead to skin breakdown and pressure injuries, especially as these devices are worn for hours at a time. If the devices do not fit securely, the air that is being pushed into the airways may instead be pushed out from the edges of the device. Patients have reported that air leaks have led to severe dry eye as the air leaks from the top of the mask and is pushed up into their eyes rather than their airway (Fauroux, et al., 2020; Knebel, et al., 1997). Similarly, there have been patient reports of nasal passage drying as the air is continuously pushed in through the nose. Oxygen delivered through the nose often requires humidification to prevent drying, possible epistaxis, and discomfort. Nevertheless, nasal drying has often been reported as a barrier to NIV usage (Cammarota, et al., 2022; Knebel, et al., 1997). Each of these factors fit into the category of discomfort, which has been the primary reported reason for failing to adhere to NIV regimens.

Another commonly reported barrier is impaired communication resulting from constant oxygen flow and placement of masks. The oxygen systems are loud which makes hearing other people and oneself difficult. Additionally, the masks make getting enough air out to formulate sentences without stopping for breaths difficult (Britton, et al., 2019). This lack of ability to communicate has resulted in increased frustration and decreased adherence to NIV.

Impaired sleep is another frequently reported barrier to these respiratory therapies. Krivec and Caggiano (2020) and Pascoe, et al. (2019) conclude that these machines frequently disrupted sleep due to general discomfort, air leakage, and pressure intolerance. The loudness of the devices was also reported to be a sleep-disrupting factor for the families of children using NIV. Due to client and family sleep disturbances, nighttime use of prescribed NIV is often decreased.

Although research has been done exploring the experiences of adults who regularly use NIV, limited research has been performed with a focus on the experience of children using NIV for respiratory support. What is known is that there is a lack of adherence to regimens leading to a decline in patient conditions because they are not receiving the oxygen they require for respiration (Al-Subu, et al., 2023). Without the therapy, these children are not getting the oxygen they need to sustain their lives. As a result, physicians must use tracheostomies or other invasive procedures that may have other, more detrimental, and life-altering side effects.

To begin researching this problem, databases such as Cinahl, ProQuest, and PubMed were utilized. Search terms such as “pediatric patients,” “neuromuscular disorders,” and “noninvasive ventilation” were entered into the aforementioned databases along with Boolean phrases to sift through thousands of articles. Despite there being many studies and literature reviews related to the subject, very few were specifically about barriers to noninvasive

ventilation use in pediatric patients and fewer still in patients with neuromuscular disorders. Pertinent sources along with those that provided basic knowledge on the subject were chosen to use for this research. However, the overall lack of research on pediatric patients with NMDs using NIV, and barriers to that use, revealed a need to expand this knowledge in pediatric healthcare.

The aim of this study is to gain a better understanding of the barriers to pediatric NIV use through the experiences of caregivers. There is a gap in the research regarding barriers to NIV use in pediatric patients which needs to be addressed. It would be dangerous to assume pediatric patients experience the same barriers as those faced by adult patients using respiratory support. Further research is necessary to better understand NIV experiences in children. We hope to understand what encourages caregivers and patients to follow their respiratory regimens and what makes following these regimens more difficult.

Methods

To explore the research objective, we could use a variety of methods: a quantitative approach, a phenomenological approach, or a qualitative descriptive approach. Each approach must be explored to determine which is the most appropriate for our research goal.

Quantitative research deals with numerical data meaning that any information gathered would have to be either measurable numerically or simple enough responses to be equated with a numerical value (for example yes = 1, no = 2). This data is valuable in assessing cause and effect relationships, hypotheses, and proposed outcomes (Carr, 1994). Simon, et al. (2012) uses quantitative methods to assess CPAP adherence in children in combination with a Likert scale

questionnaire to assess barriers to CPAP usage. Quantitative methodology would show adherence rates, oxygenation levels denoting treatment effectiveness, etc. but without the addition of a Likert scale, a questionnaire, or similar additional research method this strategy would not provide us with barriers as described by NIV users and their caregivers. Using predetermined answers (such as those in a Likert scale) would not allow study participants to answer freely and may not capture the actual specific barriers of the population. Predetermined answers to a Likert or similar scale would have to be inspired by those in an adult study which would produce biased results because we would not know if there are barriers unique to children not seen in adult participants. Our goal is to understand the experiences of these caregivers with a focus on barriers to use. This is an experience that requires too much description to be confined to the predetermined answers of a Likert scale based on previous work in adults. To obtain an in-depth understanding of pediatric NIV use, we need the words of their caregivers to more fully explain the numbers seen in other research. This is where we turn to qualitative approaches.

A qualitative study method has the potential to give us an illustration of the lives of these individuals and how their experiences affect their health. Two types of qualitative research could give us descriptions of one's life and lived experiences: phenomenology and qualitative description. Phenomenology is the study of lived experiences (Neubauer, et al., 2019). A phenomenological approach to nursing research allows us to gain a fuller understanding of one's experience as it is described in their own words (Jasper, 1994). While this is extremely valuable in obtaining a fuller picture and description of an individual's experiences and their perception of those experiences, the researcher may not necessarily gain every bit of information they wished to uncover (Wilson, 2015). For example, in phenomenology the researcher asks the participant to tell them about their experiences with a phenomena, but because the following response is the

participant's interpretations and perceptions, it is not the job of the researcher to attempt to alter or guide those with additional leading questions. While phenomenology is a valuable research methodology in nursing research, the responses we would receive from these types of interviews would not be specific enough for our purposes. By asking simply about a caregiver's experience with their child's NIV we may never learn about barriers specifically, what they are, how they have tried to work around them, or if they have experienced barriers at all. However, if we used phenomenology and asked to hear about the barriers the caregivers have experienced with their child's NIV, they may feel as though the researcher led them down a specific path or even feel blamed for not adhering to a particular treatment regimen. This could then lead to a lack of trust or comfort in sharing the fullness of their experience with NIV for their child. To answer the research question, we want to be more specific in the questions asked. We want to open the door for participants to share about every aspect of their experiences with NIV for their children, especially current and past barriers, if they have occurred, and any interventions that have or have not worked for them. Where phenomenology gives us too broad a perspective, we can use qualitative description to gain a focused understanding.

Qualitative description allows the researcher to gain firsthand knowledge about a person's perceptions of a particular event (Neergaard, et al., 2009; Sandelowski, 2000). Neergaard, et al. states, "Firstly, in terms of analysis, the aim of [qualitative description] is neither thick description (ethnography), theory development (grounded theory) nor interpretative meaning of an experience (phenomenology), but a rich, straight description of an experience or an event," (2009). By using qualitative description, the researcher can ask participants about their experiences with NIV use in their children with NMD and go into barriers, expanding as needed. This research method allows interviews to flow naturally, maintaining the trust of participants

and giving them the space to be heard. Through semi-structured interviews, the researcher would be allowed to ask the questions necessary to gain a fuller understanding of the research objectives through subsequent thematic analysis. Each of the research methods discussed have merit and can be extremely valuable in the field of nursing research, but for the purposes of this study, a qualitative descriptive approach is the most appropriate.

To reiterate, quantitative methodology would not provide us with detailed and unbiased results explaining the barriers to pediatric NIV use with NMDs. Phenomenological methodology would not allow us to ensure barriers to NIV use are discussed with the caregivers. By using a qualitative descriptive approach, we can gain an in-depth but specific description of pediatric patients with an NMD using NIV and what barriers they experience. We want to gain a full understanding of pediatric use of NIV and the barriers to regimen adherence so that we can work toward solving these obstacles and improving the respiratory statuses of these children. By doing so, we can improve their quality of life and possibly prevent the needs for tracheostomies in some cases. However, we cannot take the next steps in research without a better picture of the barriers now. To do so, we need qualitative description.

Recommendations and Discussion

There is a gap in the literature surrounding barriers to regimen adherence in pediatric patients with neuromuscular disorders. We hope to understand what encourages and inhibits caregivers and patients to follow their respiratory regimens. To understand the barriers, a qualitative descriptive approach is recommended to gain a full picture of the experiences of the caregivers. Continuing research would include demographic questionnaires and semi-structured

interviews to see what children with which NMD are using NIV, which forms of NIV they have used and are using, what barriers they may be facing, what interventions they have attempted to relieve those barriers, if the interventions have been successful, and what the caregivers' overall perceptions of NIV are for their children.

The next step would be to work with the clinical site to construct the study, identify a specific population to be interviewed, and obtain the necessary institutional approval to conduct the study. Upon completion of data collection, a thematic analysis of responses should be performed to see what, if any, themes arise from participant responses. Those themes can then be used to help address current practice issues and in future research to propose interventions to alleviate obstacles in the use of NIV in pediatric patients with NMD. By reducing the barriers to use, hopefully we can increase regimen adherence and the children's respiratory statuses as a result.

Conclusions

Quantitative, phenomenological, and qualitative descriptive approaches all have merit, but will not answer the question of barriers to NIV use in pediatric patients with an NMD. We want to learn about the experiences of the caregivers of these children with a focus on understanding the specific barriers to effective use and regimen adherence. A qualitative descriptive approach is the most appropriate method of research to understand barriers to NIV regimen adherence for children with neuromuscular disorders through the lens of their caregivers.

References

- AlBalawi, M. M., Castro-Codeal, M., Featherstone, R., Sebastianski, M., Vandermeer, B., Alkhaledi, B., Bedi, P. K., Abusido, T., & MacLean, J. E. (2022). Outcomes of long-term noninvasive ventilation use in children with neuromuscular disease systematic review and meta-analysis. *Annals of the American Thoracic Society*, 19(1), 109–119.
<https://doi.org/10.1513/AnnalsATS.202009-1089OC>
- Al-Subu, A. M., Adams, C. K., Dykstra, S. A., Langkamp, M. R., Yngsdal-Krenz, R. A., Subu, R. M. A., Zhang, Z., Baker, K. N., & Brazelton, T. B. (2023). Management of critically ill patients with spinal muscular atrophy admitted with acute respiratory failure. *American Association for Respiratory Care*, 68(2), 247-254.
<https://doi.org/10.4187/respcare.10353>
- Britton, D., Hoit, J. D., Pullen, E., Benditt, J. O., Baylor, C. R., & Yorkston, K. M. (2019). Experiences of speaking with noninvasive positive pressure ventilation: A qualitative investigation. *American journal of speech-language pathology*, 28(2S), 784–792.
https://doi.org/10.1044/2019_AJSLP-MS18-18-0101
- Brostrom, A., Nilsen, P., Johansson, P., Ulander, M., Strömberg, A., Svanborg, E., Fridlund, B., (2010). Putative facilitators and barriers for adherence to CPAP treatment in patients with obstructive sleep apnea syndrome: a qualitative content analysis. *Sleep Med*, 11, 126–130.
- Cammarota, G., Simonte, R., & De Robertis, E. (2022). Comfort during non-invasive ventilation. *Frontiers in medicine*, 9, 874250. <https://doi.org/10.3389/fmed.2022.874250>

Carr, L. T. (1994). The strengths and weaknesses of quantitative and qualitative research: what method for nursing?. *Journal of advanced nursing*, 20(4), 716–721.

<https://doi.org/10.1046/j.1365-2648.1994.20040716.x>

Diaz-Macinnis, L. (2020). Relief for air hunger: reducing severe breathlessness and psychological trauma in COVID-19 patients. *Harvard Medical School*.

<https://hms.harvard.edu/news/relief-air-hunger-0>

Ennis, J., Rohde, K., Chaput, J. P., Buchholz, A., Katz, S. L. (2012). Facilitators and barriers to noninvasive ventilation adherence in youth with nocturnal hypoventilation secondary to obesity or neuromuscular disease. *J Clin Sleep Med*, 11(12), 1409–1416.

Fauroux, B., Khirani, S., Griffon, L., Teng, T., Lanzeray, A., & Amaddeo, A. (2020). Non-invasive ventilation in children with neuromuscular disease. *Frontiers in pediatrics*, 16(8), 482. <https://doi.org/10.3389/fped.2020.00482>

Fereday, J., Muir-Cochrane, E. (2006). Demonstrating Rigor Using Thematic Analysis: A Hybrid Approach of Inductive and Deductive Coding and Theme Development. *Int J Qual Methods*, 5(1):80-92.

Green, E. & Bernoth, M. (2020). The experiences of nurses using noninvasive ventilation: An integrative review of the literature. *Australian Critical Care*, 33, 560-566.

Hansen-Flaschen, J., & Ackrivo, J. (2023). Practical guide to management of long-term noninvasive ventilation for adults with chronic neuromuscular disease. *Respiratory care*, 68(8), 1123-1157. <https://doi.org/10.4187/respcare.10349>

- Jasper, M. A. (1994). Issues in phenomenology for researchers of nursing. *Journal of advanced nursing*, 19(2), . <https://doi.org/10.1111/j.1365-2648.1994.tb01085.x>
- Katz, S., Kirk, V., MacLean, J., Bendiak, G., Harrison, M., Barrowman, N., Hoey, L., Horwood, L., Hadjiyannakis, S., Legault, L., Foster, B., & Constantin, E. (2020). Factors Related to Positive Airway Pressure Therapy Adherence in Children with Obesity and Sleep-Disordered Breathing. *Journal of Clinical Sleep Medicine*, 16(5), 733–741. <https://doi.org/10.5664/jcsm.8336>
- Knebel, A., Allen, M., McNemar, A., Peterson, A., Feigenbaum, K. (1997). A guide to noninvasive intermittent ventilatory support. *Heart Lung*, 26(4), 307-316.
- Krivec, U., & Caggiano, S. (2020). Noninvasive ventilation in palliative care and ethical dilemma. *Frontiers in pediatrics*, 8, 483. <https://doi.org/10.3389/fped.2020.00483>
- Neubauer, B. E., Witkop, C. T., Varpio, L. (2019). How phenomenology can help us learn from the experiences of others. *Perspective on Medical Education*, 8(2), 90-97.
- Neergaard, M. A., Olesen, F., Andersen, R. S., Sondergaard, J. (2009). Qualitative description - the poor cousin of health research? *BMC Med Res Methodol*, 9(52).
- Ownei, C., Rashid, S., Goswami, R., Treece, J., Shipley, L. C., Se Souza, R., O'Neill, L., Simberloff, T., & Baumrucker, S. J. (2020). Cuirass ventilation: An alternative home-based modality for chronic respiratory failure. *Home Health Care Management & Practice*, 32(1), 40–44.

- Pascoe, J. E., Sawnani, H., Hater, B., Sketch, M., & Modi, A. C. (2019). Understanding adherence to noninvasive ventilation in youth with Duchenne muscular dystrophy. *Pediatric pulmonology*, 54(12), 2035-2043. <https://doi.org/10.1002/ppul.24484>
- Popat, B., & Jones, A. T. (2012). Invasive and non-invasive mechanical ventilation. *Medicine (Abingdon, England : UK ed.)*, 40(6), 298–304.
- Prashad, P. S., Marcus, C. L., Maggs, J., Stettler, N., Cornaglia, M. A., Costa, P., Puzino, K., Xanthopoulos, M., Bradford, R., Barg, F. K. (2013). Investigating reasons for CPAP adherence in adolescents: a qualitative approach. *J Clin Sleep Med*, 9(12), 1303-1313.
- Sandelowski, M. (2000). Focus on research methods: whatever happened to qualitative description? *Research in Nursing and Health*, 23, 334–340.
- Schroth, M. K. (2009). Special considerations in the respiratory management of spinal muscular atrophy. *Pediatrics (Evanston)*, 123(Supplement_4), S245–S249.
<https://doi.org/10.1542/peds.2008-2952K>
- Simon, S. L., Duncan, C. L., Janicke, D. M., Wagner, M. H. (2012). Barriers to treatment of paediatric obstructive sleep apnoea: development of the adherence barriers to continuous positive airway pressure (CPAP) questionnaire. *Sleep Med.*, 13(2):172–177.
- Wilson, A. (2015). A guide to phenomenological research. *Nursing Standard.*, 29(34), 38–43.
<https://doi.org/10.7748/ns.29.34.38.e8821>