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Autism and neurodevelopmental disability risks in children with tracheostomies and ventilators

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Abstract

Background/Objective: Infants who survive prematurity and other critical illnesses and require continued invasive mechanical ventilation (IMV) postdischarge (at home) are at high risk of developmental delays and disabilities. Studies of extremely preterm cohorts (<28-week gestation) demonstrate rates of 25% for intellectual disability (ID) and 7% for autism spectrum disorder (ASD). Rates of ASD and ID in children with IMV are unknown. This study aimed to determine neurodevelopmental disability risk in a cohort of children with postdischarge IMV.

Design/Methods: A consecutive series of children with IMV were assessed 1 month, 6 months, and 1 year after discharge. Cognitive, social, and communicative domains were assessed by a Developmental and Behavioral Pediatrician using (1) clinical adaptive test/clinical linguistic and auditory milestone scale (CAT/CLAMS) of the capute scales; (2) pediatric evaluation of disability inventory computer adaptive test (PEDI-CAT); and (3) modified checklist for autism in toddlers, revised (MCHAT-R). Red flag signs and symptoms of ASD using DSM-V criteria were noted. Longitudinal testing was reviewed. Expert consensus impressions of evolving ASD and/or ID were determined.

Results: Eighteen children were followed for 1 year; at 1 year, the median age (range) was 23 (17–42) months. Children were 44% male, 33% non-Hispanic White, 39% non-Hispanic Black, and 28% Hispanic. Fifteen (83%) children were prematurity survivors. Median (range) developmental quotients (DQs): full-scale DQ 59 (11–86), CAT DQ 66.5 (8–96), and CLAMS DQ 49.5 (13–100). Twelve (67%) children were highly suspicious for ASD and/or evolving ID.

Conclusions/Significance: This cohort of children with at-home IMV demonstrates a higher risk of ASD and ID than prior premature cohorts. Larger investigations with longer follow-up are needed.

KEYWORDS
childhood development, childhood disability, children with medical complexity, children with ventilator dependency

Robert J. Graham and Michael E. Msall contributed equally to this study as senior authors. An abstract of this work was presented at the American Academy of Cerebral Palsy and Developmental Medicine 2023 meeting.

1 | INTRODUCTION

Infants who survive prematurity and other critical illnesses but continue to require invasive mechanical ventilation (IMV) assistance beyond hospital discharge are at high risk of developmental delays and disabilities. Studies of extremely preterm cohorts (<28 weeks' gestation) demonstrate rates of 25% for intellectual disability (ID)[1] and 7% for autism spectrum disorder (ASD)[2] at 10 years of age. In a cohort study across 16 centers of the Eunice Kennedy Shriver National Institute of Child Health and Human Development Neonatal Research Network, De Mauro et al. identified that toddlers with a history of tracheostomy had substantially higher odds of death or neurodevelopmental impairment when compared to similarly premature children.[3] This increased vulnerability of children with IMV may be due to unique developmental challenges, including underlying conditions, acquired injury from periods of cardiorespiratory instability, prolonged hospitalizations during critical developmental periods, and limitations on early expressive speech and feeding.

ID is defined as significant limitations in both intellectual functioning (cognitive skills in perception, memory, learning, reasoning, problem-solving) and adaptive behavior (conceptual, social, and practical skills that are learned and performed by people in their everyday lives) that originates before the age of 22.[4] It is estimated that children born preterm have at least twice the risk of having ID; children born at 27 weeks may be as high as eight times as likely to have ID when compared with term-born peers, for a total population prevalence of 5.6%.[5] Overall, children born extremely preterm or very preterm have IQ scores 13 points lower than term peers; there is evidence that this relationship is linear.[6] The rates of ID in children with IMV are largely unknown.

ASD is a complex neurodevelopmental disorder defined as persistent deficits in each of the three areas of social communication and interaction: social-emotional reciprocity, nonverbal communicative behaviors used for social interaction, and relationships, as well as restrictive, repetitive behaviors and interests, and hyper or hyporeactivity to sensory input.[7,8] Children born prematurely,[9] born small for gestational age,[10] or those with peripartum stroke or intracranial bleeding are established as having higher rates of ASD. The rates of ASD in neonates with IMV, however, are largely unknown.

Due to overlapping phenotypic characteristics, particularly in early childhood, in some cases, the diagnoses of ASD and ID are difficult to differentiate.[11] In clinical practice children are often older before formally receiving a diagnosis of ASD, ID, or both. ASD is typically not diagnosed before age 12 months, which coincides with the Autism Diagnostic Observation Schedule threshold,[12] and ID is typically not diagnosed until later childhood when IQ can be combined with measures of adaptive functioning.[13] In this study, we sought to use expert clinical longitudinal developmental assessments in a prospective neonatal cohort of children with IMV to determine overall risks for severe neurodevelopmental disability, including those with only concerns for ID, only concerns for ASD, and concerns for a dual diagnosis of ID and ASD.

2 | METHODS

2.1 | Participant recruitment and study methods

Participants were recruited through The University of Illinois at Chicago Division of Specialized Care for Children (DSCC) Home Care Program. Eligible participants included children (<18 years of age) with new IMV transitioning home from the hospital for the first time. Long hospitalizations can impede developmental progress.[14] This study was designed to use expert evaluation to track an index cohort of children with home ventilation to understand the patterns of developmental gains in socially stimulating home and community settings. The study visits by the clinical research coordinator (Emma Lynch) and Developmental and Behavioral Pediatrician (Sarah A. Sobotka) including sequential developmental assessments were for the purposes of this research study only and are not standard clinical practice. For this analysis, we focused on developmental skills after neonatal illness and we excluded three child participants who had ventilation beginning after infancy. Details of the methodology have been described in prior published work.[15] This study was approved by the University of Chicago Institutional Review Board (IRB17-0908). Strengthening the reporting of observational studies in epidemiology (STROBE)[16] guidelines were used to report developmental testing findings.

2.1.1 | Follow-up developmental assessments

Enrollment developmental testing was completed 1 month after hospital discharge.[15] Written consent in English or Spanish was obtained for full study participation including sequential developmental testing, parent interviews, and data extraction from the DSCC letter of medical necessity. Follow-up assessments occurred 6 months and 1 year after enrollment. Two investigators (Sarah A. Sobotka and Emma Lynch) participated in all follow-up family home visits. The 6-month follow-up study visits comprised three components: (1) An expert developmental assessment; (2) Parent survey-based developmental assessment tools; and (3) An interview with parent(s). The interview included topics related to habilitative services and developmental outcomes. Therapies received at this 6-month time period were determined from the analysis of these interviews. The 1-year follow-up study visit included developmental assessments only. An in-person Spanish translator was utilized for primarily Spanish-speaking families. Visits after March 2020 were temporarily converted to Zoom conferencing and often split into two visits due to length. After August 2020, families were given the choice of in-person or video. Nine 6-month follow-up visits and nine 1-year visits were conducted completely or partially via video conference. The median (range) time between enrollment and 6-month follow-up was 6 (5–6) months ($n = 16$), and the median time between enrollment and 1-year follow-up was 12 (11–14) months ($n = 18$). Two 6-month follow-up visits were missed due the

child being hospitalized for extended periods; both subjects re-engaged at 1 year, and interviews were completed at that time.

2.1.2 | Interactive developmental assessments

Developmental testing was completed by a subspecialty-trained Developmental and Behavioral Pediatrician (Sarah A. Sobotka) to understand children's strengths and challenges in mobility, changing positions, manipulating objects, expressive and receptive communication, integrating vision for learning, and performing basic self-care skills. The clinical adaptive test/clinical linguistic and auditory milestone scale (CAT/CLAMS), also known as the Capute scales, enables a comparison of language-related and nonlanguage-related problem-solving skills to aid in diagnosing developmental delay and disability.[17] The CAT/CLAMS is designed to correct for gestational age. Corrected developmental quotients were calculated for all children born prematurely under 2 years of chronological age.[18] Performance at 24 months of biological age and beyond was no longer corrected. Developmental quotients quantify differences from age-appropriate performance (e.g., a DQ of 50 indicates performance at 50% below average, such as having the skills of a 6-month-old when the corrected gestational age is 12 months).

2.1.3 | Parental survey-based developmental assessments

Parental assessments included the pediatric evaluation of disability inventory computer adaptive test (PEDI-CAT), which measures the child's daily activities, mobility, and social/cognitive abilities, thus assessing the child's ability to manage complex life tasks.[19] Parents of children aged 16 months and older also completed the modified checklist for autism in toddlers, revised (MCHAT-R), which is a standardized screening tool used to screen for ASD.[20]

2.1.4 | Demographic survey and record review

Upon enrollment, a demographic survey was completed by parents using the research electronic data capture system (REDCap).[21] Clinical data were extracted from the DSCC letter of medical necessity or service renewal letter.[15] Neighborhood socioeconomic disadvantage was measured by the National Area Deprivation Index, which we calculated using a 9-digit zip code.[22,23]

2.1.5 | Statistical analysis

For analysis of the PEDICAT, Stuart-Maxwell (marginal homogeneity) tests were completed for all comparisons in subscales and between time point analysis. For the Capute Scales, the Shapiro–Wilk test was used to assess for normal distribution, and paired *T*-tests were used to compare responses between time points. Data with a normal distribution are presented as mean and standard error. All comparisons between study time points were made with Bonferroni corrections to consider *p*-values significant if <0.05.

3 | RESULTS

Twenty children with neonatal illness requiring IMV were enrolled in the study. After initial enrollment participation, one child died before follow-up, and one had a catastrophic anoxic brain injury; these children were excluded from this longitudinal analysis. Between February 2019 and September 2022, 18 children who had survived neonatal critical illness with IMV assistance were followed for three study visits over a median (range) 12 (11–14) months. Eighteen children were assessed at a median age (range) of 23 (17–42) months old; 44% male, 33% non-Hispanic White, 39% non-Hispanic Black, and 28% Hispanic. Fifteen (83%) children were survivors of prematurity, and five (28%) had a genetic disorder. The median (range) National Area Deprivation Index was 40 (6–96) (Table 1).

Caregivers reported 10 (56%) children were receiving developmental therapy, eight (44%) were receiving feeding therapy, eight (44%) were receiving nutrition services, 14 (78%) were receiving occupational therapy, 16 (89%) were receiving physical therapy, and 11 (61%) were receiving speech therapy. The majority of follow-up interviews (94%) happened after the start of the COVID-19 pandemic; 83% of these reported that the pandemic affected therapy services. Families described delayed assessments, conversion to virtual therapies, and a lack of services entirely (Table 2).

3.1 | Developmental outcomes

PEDICAT: All subscales were compared between time points. At each time point, the most common pattern was no change in percentiles, with the percentile of function being either <5% or 5%–25%. With Bonferroni correction, no comparisons between time points of any time scale were significantly different. Cohort distributions are displayed in Table 3 and Figure 1.

Capute scales: Median (range) CAT DQ was 62.5 (3–113) at enrollment, 70.5 (8–106) at 6 months, 52.5 (12–116) at 1 year. Median (range) CLAMS DQ was 74.5 (11–125) at enrollment, 56.5 (13–100) at 6 months, and 53 (11–98) at 1 year. Individual patterns of performance over time are displayed in Figure 2A,B. Pairwise Comparisons of CAT and CLAMS subscales at each time point are displayed in Table 4. Enrollment CLAMS versus 1-year CLAMS showed significant worsening over time with a *p*-value of 0.041. The nonsignificant comparisons demonstrate no change in developmental quotient (i.e., no catch-up development) over the observation period.

ASD/NDD clinical assessment: Children were evaluated in the family home or over video conferencing three times during the study by a Developmental and Behavioral Pediatrician (Sarah A. Sobotka).

TABLE 1 Enrollment characteristics of children with invasive mechanical ventilation and their parents ($n = 18$).

Demographic Characteristics	*n* (%)
Child characteristics	
Male child	8 (44)
Race/Ethnicity of Child	
Non-Hispanic White	6 (33)
Non-Hispanic Black	7 (39)
Hispanic	5 (28)
Age of child at 1-year follow-up in months median (range)	23 (17–42)
Diagnoses	
Prematurity	
Extreme (<28 weeks)	10 (56)
Very (28–32 weeks)	2 (11)
Moderate to Late (32–37 weeks)	3 (17)
Term	3 (17)
Congenital heart condition	9 (50)
Genetic disorder	
Duplication or deletion	3 (17)
Another genetic anomaly	2 (11)
Hearing loss	6 (33)
Retinopathy of prematurity	9 (50)
Number of subspecialists (mean [range])	7 (3–12)
Medical Equipment	
Feeding tube	18 (100)
Tracheostomy	18 (100)
Ventilator	18 (100)
Hospitalization characteristics	
Length of hospital stay (days median [range])	303 (132–788)
Admission included transitional care center	8 (44)
Parent/family characteristics	
Enrolled parent's relationship to child[a]	
Mothers participated alone	7 (39)
Father and mother interviewed together	11 (61)
Marital status	
Married	12 (67)
Single	3 (17)
Other	3 (17)
Number of other children in the household	
0	3 (17)
1	6 (33)
2	7 (39)
3	2 (11)
Household income <$50,000 ($n = 17$)	9 (53)
National Area Deprivation Index median (range)	40 (6–96)
Work status	
Full-time or part-time	7 (39)
Not currently working	11 (61)
Change in work status due to care of child	
No change in work hours	6 (33)
Decrease in work hours	12 (67)

[a]Both parents were encouraged to participate if available; answer reflects parent completing survey.

TABLE 2 Received therapy services for cohort at 6 months follow-up ($n = 18$).

Therapy service	*n* (%)
Receiving early intervention services	
Developmental therapy	10 (56)
Feeding therapy	8 (44)
Nutrition services	8 (44)
Occupational therapy	14 (78)
Physical therapy	16 (89)
Speech therapy	11 (61)
EI services affected by COVID-19	15 (83)

Red-flag signs and symptoms for ASD were noted during the longitudinal developmental assessments and notes taken for future case discussions. For example, the examiner would document a child's lack of eye contact with the parent and examiner, complex finger mannerisms, or self-injurious behaviors. Data from each complete neurodevelopmental evaluation for each child were discussed in consultation with another neurodevelopmental expert (Michael E. Msall). Evolving ID was considered for any child with a full scale DQ < 70% at 1-year follow-up, paralleling the intellectual quotient thresholds for consideration of ID.[8] Each child's developmental assessments were reviewed to determine whether or not they were highly suspicious for ASD and/or evolving ID. For example, a child with full scale DQ of 31 was determined to have evolving ID. Another child with a full-scale DQ of 65, met the criteria based on IQ but was also noticed to exhibit looking out of the corner of their eye, hand flapping, and hitting their head. Consensus opinion determined that 12 (66.7%) children were highly suspicious for ASD and/or evolving ID. Of note, these children categorized as evolving ASD/ID were all at least 21 months of age at their 1-year assessment. Gold standard assessment for a diagnosis of ASD using the Autism

TABLE 3 The pediatric evaluation of disability inventory computer adaptive test (PEDI-CAT) results for cohort ($n = 18$).

Score			Participants' enrollment outcome $n = 18$ n (%)	Participants' 6 month outcome $n = 16$ n (%)	Participants' 1 year outcome $n = 18$ n (%)
Subscore	Daily activities percentile	<5%	14 (78)	6 (38)	7 (39)
		5%–25%	3 (17)	8 (50)	7 (39)
		25%–50%	0	1 (6)	1 (6)
		>50%	1 (6)	1 (6)	3 (17)
	Mobility percentile	<5%	13 (72)	8 (50)	15 (83)
		5%–25%	4 (22)	6 (38)	1 (6)
		25%–50%	1 (6)	1 (6)	1 (6)
		>50%	0	1 (6)	1 (6)
	Social/cognitive percentile	<5%	7 (39)	5 (31)	7 (39)
		5%–25%	9 (50)	8 (50)	8 (44)
		25%–50%	1 (6)	3 (19)	2 (11)
		>50%	1 (6)	0 (0)	1 (6)
	Responsibility percentile	<5%	6 (33)	10 (38)	12 (67)
		5%–25%	10 (56)	6 (63)	2 (11)
		25%–50%	1 (6)	0 (0)	1 (6)
		>50%	1 (6)	0 (0)	3 (17)

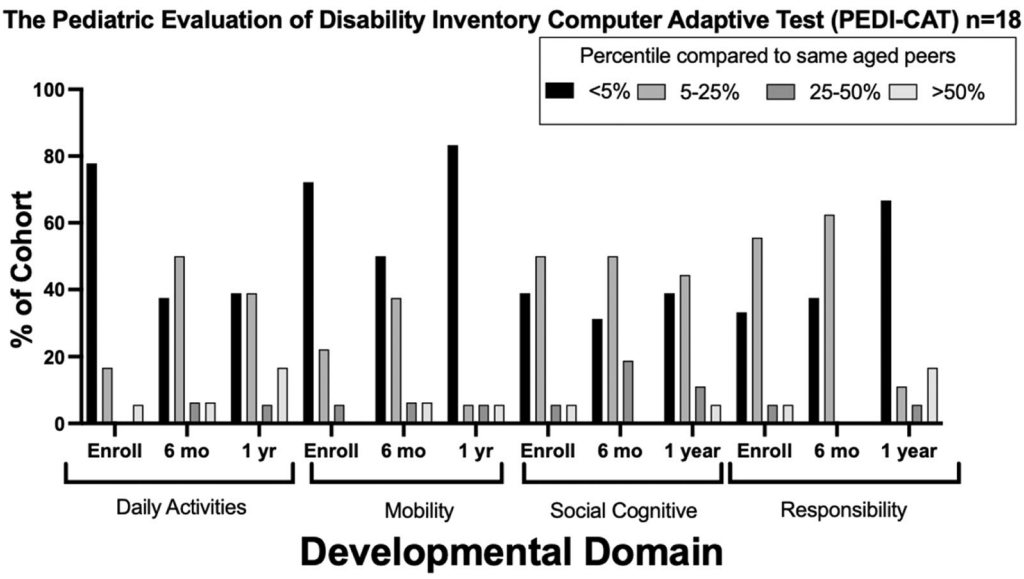

FIGURE 1 The pediatric evaluation of disability inventory computer adaptive test (PEDI-CAT) $n = 18$.

Diagnosis Observation Scale—Toddler Module is a standardized tool for testing for ASD in children as young as 12 months of age,[12] thus, it is within clinical practice to consider an ASD diagnosis in toddlers. Some children who were not categorized in this analysis as highly suspicious for ASD and/or evolving ID still had at least one concerning sign or symptom of ASD. For example, a child with a CAT DQ of 70 and low-risk MCHAT at 1 year, was observed to have finger mannerisms and become upset with loud noises, and was categorized as not highly suspicious. In clinical practice, this would warrant close follow-up and re-evaluation for ASD. In this analysis the investigator team opted for a conservative threshold for ASD/ID categorization.

4 | DISCUSSION

This pilot cohort of children requiring IMV at the time of NICU discharge demonstrated a higher risk of evolving neurodevelopmental disability, ASD and/or ID, than prior estimates of premature cohorts. Sequential neurodevelopmental testing over more than a year after initial hospital discharge did not demonstrate developmental catch-up, as is often expected by parents and providers. Rather, performance compared to age-matched cohorts remained relatively static, with a notable worsening of linguistic milestones.

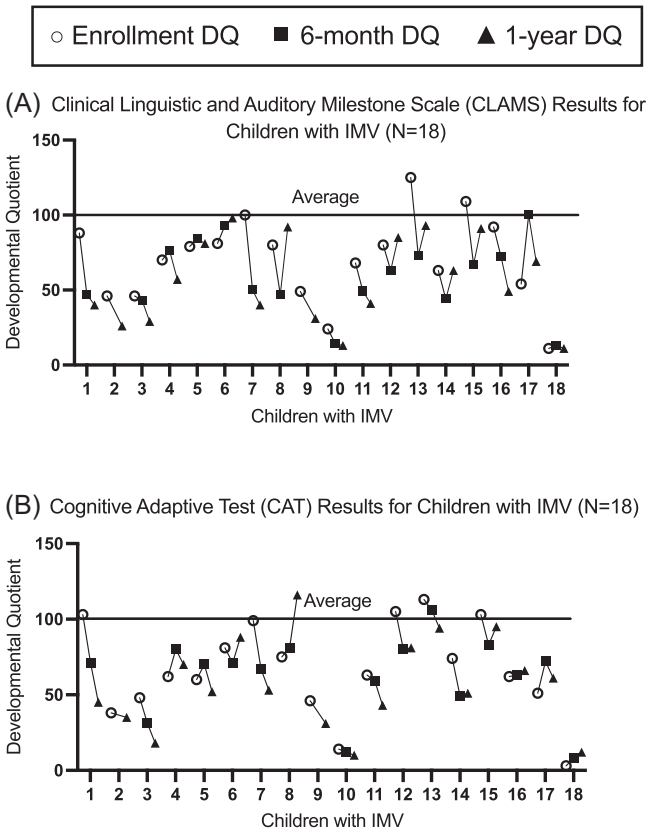

FIGURE 2 Capute scale results at enrollment, 6-months, 1 year (n = 18).

We interpret these findings as suggestive cautionary evidence, yet not entirely pessimistic findings. Over time, children with IMV continue to grow and learn new developmental skills, especially when supported by caregivers in stimulating environments with guidance from expert therapists. However, many providers and parents have expectations for catch-up development, which would require children with IMV with developmental delays to learn at a *higher rate* than typically performing peers. Although cautious not to overgeneralize about this population based on our small sample, we interpret our findings of relative lack of developmental change over 1 year postdischarge as a challenge to the expectation for developmental catch-up. This difference between *learning* and *rate of learning* is an important distinction that should be communicated when discussing developmental expectations with families.

Perhaps most intriguing is the high suspicion rate for ASD in the study cohort. We note that prior estimates of ASD among high-risk premature cohorts were far lower, 7%.[2] We consider three possible explanations for this surprising finding. First, our small sample may be unrepresentative of the population of children with IMV and through chance more skewed toward a cohort of children with ASD. Second, children with IMV are children with neonatal illness who experienced more severe critical medical illness and extreme cardio-pulmonary vulnerability requiring ongoing support beyond hospitalization. These children may also have increased overall vulnerabilities to their neurodevelopment. Third, children with IMV experience long hospital stays, which separate children from typical social engagement opportunities, including parent-child, sibling, and peer interaction and often are primarily characterized by time alone in a crib.[24] Further, overall sensory experiences during hospitalizations differ from typical infancy dramatically and may influence sensory responsiveness. These environmental impacts may affect ASD risk.

This study should be interpreted with caution in light of important limitations. This small cohort of 18 children with neonatal and postdischarge IMV is not representative of either the heterogeneity nor the average performance of the IMV population. While strategies were employed to procure an unbiased sample (recruiting through a

TABLE 4 Pairwise comparisons of clinical adaptive test/clinical linguistic and auditory milestone scale (CAT/CLAMS) at different time points by paired T-test.

Pair	N	Mean (Standard error)	p-Value[a]
Enrollment CAT versus 6-month CAT	16	69.8 (7.9) versus 62.7 (6.5)	0.346
Enrollment CAT versus 1 year CAT	18	66.7 (7.3) versus 56.7 (7.1)	0.245
6-month CAT versus 1 year CAT	16	62.7 (6.5) versus 59.7 (7.7)	>0.999
Enrollment CLAMS versus 6-month CLAMS	16	73.1 (7.4) versus 58.4 (6.2)	0.117
Enrollment CLAMS versus 1 year CLAMS	18	70.3 (6.8) versus 56.1 (6.8)	0.041[b]
6-month CLAMS versus 1 year CLAMS	16	58.4 (6.2) versus 59.5 (7.2)	>0.999

[a]After Bonferroni correction.

[b]Significant at <0.05 level.

state-wide organization), and the cohort was notably racially and socioeconomically diverse, there may be unintended biases in small samples. We note that our sample had a low national area deprivation index as compared to national mean, and we appreciate the important impacts of family and neighborhood socioeconomic disadvantage on child development and disability.[25] We also acknowledge that therapies have the potential to influence outcomes, yet in our cohort children were not universally receiving a full cadre of habilitative therapies. We encourage future investigations with larger populations and geographic distributions which would enable multivariable analysis including environmental, genetic, and health service risk factors, to improve our understanding of contributory risk factors. We also acknowledge that gold standard diagnostic testing for ASD and ID was not completed, and for many could not have because of their young age. However, during a period of time immediately after hospital-to-home transition when many experts and parents might have expected catch-up in developmental skills, our study demonstrates maintained delays with frequently widening gaps.

We also acknowledge our lack of precision by deciding to group together risk of ID and ASD. In this young cohort, we chose to combine risks of ID and ASD to identify an overall likelihood of neurodevelopmental disability requiring intensive habilitative and educational supports. Further, we acknowledge that ID and ASD have both genotypic and phenotypic overlap, especially in early childhood.[11] A longitudinal study of special education records demonstrated that diagnoses can switch or broaden to dual diagnoses over early and late childhood.[26] Therefore, although we encourage further studies to complete comprehensive neuropsychological testing in later childhood to more precisely estimate neurodevelopmental risk, the combination of ID and ASD in this cohort as indicators of neurodevelopmental disability achieved the study goal.

This study has important implications for future studies and clinical innovations. First, hospital-based providers who intersect with this population ought to consider the higher neurodevelopmental risk in this cohort when counseling families on future expectations. This counseling should be sensitive to the unknown etiologies of disabilities; IMV may likely be a marker for neurodevelopmental risk rather than causal, although we acknowledge that the presence of the tracheostomy and ventilator inhibits some aspects of developmental exploration. Most importantly, providers sensitive to these increased vulnerabilities must work to mitigate risk and maximize developmental potential. We note that our cohort did not universally participate in broad habilitative therapies and many gaps existed. Access to habilitative interventions is even more challenging for this often-homebound population, yet needs are vast, and for many worsened in the setting of the COVID-19 pandemic. Prior studies have demonstrated that children with IMV do not receive community-based therapies,[15] in part their dependence on medical technologies may intimidate therapy and early intervention providers.[27] Additionally, long delays from hospital-to-home despite children being medically ready for discharge[28,29] means that community-based therapies are further postponed. Programs which expedite habilitative services once the children are finally in the home setting would best serve this neurodevelopmentally vulnerable population.

5 | CONCLUSION

Children with home IMV with onset in the neonatal period may have higher rates of ID and ASD than previously estimated. These high-risk profiles highlight the need for studies which follow children into later childhood and determine precise estimates of neurodevelopmental disabilities. Most importantly, tailored therapeutic strategies for children with IMV are needed to optimize neurodevelopmental outcomes in this vulnerable population at high risk for major disability.

AUTHOR CONTRIBUTIONS

Sarah A. Sobotka: Conceptualization; investigation; funding acquisition; writing—original draft; methodology; writing—review and editing; formal analysis; supervision; project administration; visualization. **Emma Lynch**: Writing—review and editing; project administration; data curation. **Chuanhong Liao**: Formal analysis; software; writing—review and editing. **Robert J. Graham**: Writing—review and editing. **Michael E. Msall**: Writing—review and editing; conceptualization.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

This study was approved by the University of Chicago Institutional Review Board (IRB17-0908) and informed consent was obtained from all participants.

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