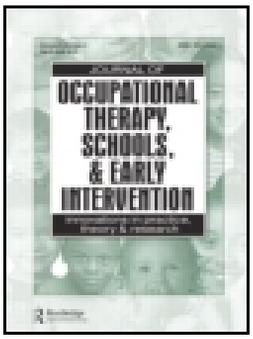

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Motor Milestones: Sensory Motor Trends of Young Children with Classic Galactosemia

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ABSTRACT

Speech problems affect about 66% of children with classic galactosemia (CG), but limited evidence is reported on early motor and sensory motor development in this at-risk population. Research has been focused on speech and language development, leaving a paucity of data on motor and sensory differences. This paper describes preliminary data regarding sensory motor and motor development patterns in young children with CG. Babble Boot Camp© (BBC) is an NIH-funded randomized control trial (RCT) implementing proactive interventions designed to support the speech language development of infants with CG. Cases were randomly assigned to a motor-first group (Motor Milestones), receiving virtual occupational therapy through 14 months, or a speech-first group, receiving virtual speech therapy through 14 months. All cases received speech and language therapy from 15 to 24 months. Controls, typically developing infants, did not receive occupational therapy or speech therapy. Participants were recruited through social media, advertisements, metabolic clinics, and the Galactosemia Foundation. Infants in the motor milestones group were assessed with the Developmental Assessment of Young Children and Sensory Profile-2 pre-enrollment (<6 months of age) and post-treatment follow-up at 2.5, 3.5, and 4.5 years of age. Results show that 17.5% of participants with CG had delays in gross motor, 22.5% in fine motor, and 45% in sensory processing. Data from the Motor Milestones portion of BBC are important emerging evidence for occupational therapy in early intervention, preschool, and outpatient settings. This research supports the need for occupational therapy services during early intervention to minimize or prevent long-term motor and sensorimotor delays in infants with CG. Understanding patterns and addressing literature gaps helps support the need for occupational therapists to address motor delays, improve activities of daily living, play, promote functional independence, and provide caregiver education to best support the occupational performance of children with CG.

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Background

Classic galactosemia (CG) is a rare genetic inborn error of metabolism diagnosed via newborn screening. CG leads to the inability to break down the sugar galactose, typically found in dairy, breast milk, and other foods. Diagnosis is determined when newborn screening detects a significant deficiency in erythrocyte galactose-1-phosphate uridyltransferase (GALT) activity (Fridovich-Keil & Berry, 2022). CG is rare and differs by geographic location. The reported incidence rates of CG in Western countries range between 1 of 16,000 and 1 of 60,000 live births, with highest incidence rates among individuals of Irish descent (Rubio-Gozalbo et al., 2019). Although diet restrictions are implemented immediately upon detection, thus avoiding diet-induced serious illness or death, children with CG have long-term developmental complications (Berry, 2000; Fridovich-Keil & Berry, 2022). Approximately 52% of children with CG have global developmental delays, 27% demonstrate motor abnormalities, and 66.4% have a speech-language disorder (Rubio-Gozalbo et al., 2019). Detailed reporting on motor differences and a total lack of reporting on sensory processing patterns are limits to the evidence for children and families with CG. Research utilizing functional MRI imaging of individuals with CG found that there is altered connectivity to the frontal gyrus and insular cortex, which is important for sensorimotor integration and motor planning (Van Erven, Jansma, Rubio-Gozalbo, & Timmers, 2017). Understanding the differences in structural connectivity in addition to the preliminary data outlined in this study provides evidence regarding the prevalence of sensory processing in young children with CG.

Babble Boot Camp© began initially as a speech and language intervention study in partnership with speech and language pathologists (Peter et al., 2021). Not only at risk for speech and language delays, children with CG also have marked delays in fine and gross motor development (Berry, 2000; Fridovich-Keil & Berry, 2022). Currently, limited evidence exists regarding the prevalence of children with CG and sensorimotor delays. To address the dearth of evidence, the team expanded the scope of Babble Boot Camp© to also provide support to participants and their families for sensory motor and motor development. Motor Milestones, a proactive intervention focusing specifically on development of fine motor, gross motor, and sensory processing, were created for families who were randomized into the later speech intervention. Motor Milestones teleconference sessions were conducted bimonthly by an occupational therapist in an effort to provide consistent care for infants with CG.

A developmental profile of children with CG helps occupational therapists better understand this population and requires occupational therapists to become more active participants in making sure children with CG achieve their developmental milestones. By understanding the developmental trends of infants with CG, a preventative and proactive health approach can be taken during the evaluation and treatment of infants with CG. There is currently limited evidence on sensory processing differences and motor development for children with CG; thus, this study addresses this gap and explores findings that are relevant to practitioners in schools and EI settings. These data are presented with the aim of improving the evaluation and treatment of CG across the world, particularly to share developmental data relevant to occupational therapists in early intervention (EI) and school-based practice (SBP). This study presents the preliminary findings of an ongoing research study, Babble Boot Camp© (Peter et al., 2019, 2021), designed to proactively

support the development of infants with CG to address language delays and establish the empirical evidence regarding sensory-motor development. In particular, this manuscript seeks to answer the following research questions related to the proportion and normalized distribution of atypical development in CG infants relevant to occupational therapists and multi-disciplinary teams found in EI and SBT settings. 1) What are sensory processing patterns in children with CG? 2) What are the developmental motor patterns of children with CG? 3) What are the developmental patterns of adaptive skills of children with CG?

Methods

This study was conducted with the oversight and approval of the XXX (redacted for peer review) Institutional Review Board. Parents provided written consent for their children to participate after thorough explanation of the study. Upon enrollment into the study at age <6 months, infants with CG (cases) were randomized into one of two treatment cohorts: Talk Time First or Motor Milestones. The Talk Time First cohort received speech/language intervention from age <6–24 months, and the Motor Milestones cohort received sensory and motor support from age <6–15 months and then speech/language intervention from 15–24 months. The authors intended to examine and establish baseline data for motor and sensory trends for children with CG through the use of DAYC and Sensory Profile-2 capturing the developmental profile of children with CG. Effectiveness of intervention will be addressed in future data analysis. From these assessments, data were collected from 40 participants. Averages were calculated using data from entrance and subsequent follow-up sessions to determine the number of participants who had atypical motor and sensory scores within the infant, toddler, and child age groups.

Participants

Participants for this study were infants living in the United States (US), United Kingdom (UK), and Canada with a newborn diagnosis of CG, <6–24 months of age. None of these participants had other subtypes of CG or medical diagnosis that could introduce confounding variables (e.g., trisomy 21, deafness). Controls were typical infants and children with CG who were older than 6 months of age when enrolled in BBC. All controls received conventional care but not BBC intervention. Controls were required to be free of CG and any condition or trait that could introduce confounding and additionally. Controls were matched to children with CG based on entry into the study prior to 6 months of age. Typical controls were recruited for development to be observed using the same tools as those used for the children with CG. At the time of this analysis, the Talk Time First cohort consisted of 25 children (13 male, 12 female) with a mean age of 13 months and an age range of 7 to 23 months. Additionally, at the the of analysis, the Motor Milestones cohort consisted of 40 participants (27 female, and 13 male) with a mean age of 6 months and an age range of 2 to 23 months.

Children with CG were recruited through the Galactosemia Foundation, social media, and word of mouth. Additionally, flyers are distributed to obstetricians, pediatricians, and day-cares. Typical controls were recruited via social media and included individuals in the US and Canada. Participants in the UK and Canada contacted the BBC team after finding the study announcement on the Galactosemia Foundation website. All participating

families in and outside the US spoke English as the primary language. Nationality or citizenship was not tracked for the purposes of this study. Of the children with CG, 17% resided in the UK and Canada. That percentage was 19 for the typical controls, so that the ratio of participants residing outside and inside the US was highly similar among the children with CG and the typical controls.

The participant fees were set at an amount that was considered below the level of inducement while also motivating families to complete their participation in the study. $100 for the CG treatment families and $50 for the control families when testing at 24 months is complete; at the last follow-up testing at child age 48 months, the lump sum of $150 is provided to incentivize families to stay enrolled until the end of the study. Families who participated in the intervention received a higher fee than the control families due to the required weekly time demands. Inclusion criteria for case participants include entry into the study at about 2 months, a newborn diagnosis of CG, English as primary language, internet and computer access, and parents must have 8th grade education or higher to fill out forms. Case and control participants were excluded who had a diagnosis of CG or other forms of CG. Additionally, none of the participants had a medical diagnosis that could introduce confounding variables (e.g., trisomy 21, deafness). This study was funded by the National Institute of Health Grant (R01 HD098253–01). Enrollment of families in the UK and Canada was approved via a formal process. The Institutional Review Board at Arizona State University provided ethical oversight. The study location for all foreign participants is the US, as the intervention was provided via telehealth. No foreign institutions needed to provide approval.

Procedure

The Motor Milestones cohort consisted of infants diagnosed with CG. Evaluation consisted of norm referenced motor, sensory, and adaptive measures. The assessments were completed virtually using a HIPAA-compatible telehealth platform, through parent report, observation, and parent interview. At entry in the study, participants are evaluated and again at conclusion of intervention at 24 months. Post-assessments of the same measures were utilized at 30, 42, and 54 months of age. Assessments are hand-scored and checked by a non-assessing individual to prevent error and establish inter-rater reliability.

Measurement Tools

Measured outcomes included metrics of motor, sensory, and adaptive development. The Developmental Assessment of Young Children (DAYC-2), and Sensory Profile-2 were used to assess infants pre- and post- the intervention period. For these assessments, normative data were collected in the US and utilized for British and Canadian participants creating a limitation in the study. This has been reported and acknowledged in the limitation section below. Developmental differences were interpreted using standard deviation from the mean (SD); developmental differences are categorized as 1 SD above or below the mean and definite difference 2 SD.

Sensory Profile-2 is a parent questionnaire, utilizing parents' reports on their child's behavioral responses to sensory stimulation. The questionnaire measures parent response to elicit sensory processing patterns. The assessment is dependent on age and broken up

into an infant, toddler, and child form. The assessment provides a score profile in four different quadrant areas: seeking, avoiding, sensitivity, and registration. Sensory seekers require additional input, so they tend to be busier and more engaged in sensory experiences. Avoiders are bothered by sensory input and are more likely to retreat from unfamiliar situations. Sensors are hyper aware of sensory input, reacting more quickly and intensely than others. Those categorized as registration may miss sensory input, missing stimuli that might be annoying to others. In addition to providing normed scores in each quadrant, Sensory Profile 2 also provides normed information for each sensory category (auditory, visual, touch, movement, body position, and oral). For the purpose of this study, this assessment was completed utilizing structured parent interviews (Dunn, 2014). Convergent validity of sensory profile revealed moderate convergent validity between the sensory profile and the sensory processing measure home form as indicated by Spearman's Rho equaling 0.86, $p < .01$ (Dunn, 2014). Test–retest reliability for all parent questionnaires ranges from .83 to .97 (Dunn, 2014). Inter-rater reliability ranges from .49 to .89 and internal consistency from .57 to .90 (Dunn, 2014).

The DAYC-2 is a norm-referenced assessment that measures development in the areas of cognition, communication, social-emotional development, physical development, and adaptive behavior. Each domain can be assessed independently. For the purpose of this study, the physical development and adaptive behavior subtests were utilized. The physical development domain involves two sub-domains of fine and gross motor development. Gross motor skills involve large muscles in arms and legs that help us walk, run, skip, and jump. Fine motor skills involve small muscles in hands and wrists that help us write, cut, or buttoning. Finally, the adaptive domain incorporates self-help skills in the area of Activities of Daily Living (ADLs). The DAYC-2 provides flexibility regarding constraints of completing assessment virtually. The assessor is able not only to directly observe skills required but also obtain parent reports for skills not easily observed virtually (i.e., walks upstairs). This provided flexibility to incorporate structured observation and parent interview when completing this assessment virtually. BBC began initially as a speech and language intervention study, with the assessments and interventions being used by the speech and language team to heavily address cognition, communication, and social emotional behavior. The occupational therapy-led portion of the study began to provide the families who were randomized into the later intervention group for speech some support in overall development. Therefore, the motor team focused primarily on physical development and adaptive behavior as these components were identified as gaps in the current data that could be filled by the occupational therapy practitioners on this research team. Reliability of the DAYC-2 was measured by evaluating three sources of error: coefficient alpha, test-retest, and scorer difference (Voress & Maddox, 2013). The reliability of the DAYC-2 presented with a coefficient alpha of .90, a test–retest value ranging from .70 to .91, and scorer difference at .99, all providing sufficient evidence of the reliable nature of the DAYC-2 (Voress & Maddox, 2013). Additionally, the DAYC-2 is a valid measure of indicating early childhood development as evidenced by 1) content description validity had distinct rational for format and content of each domain and the validity of item analysis being reinforced by results of differential item functioning analyses used to show the absence of bias in a tests' items; 2) criterion-prediction validity showed coefficients

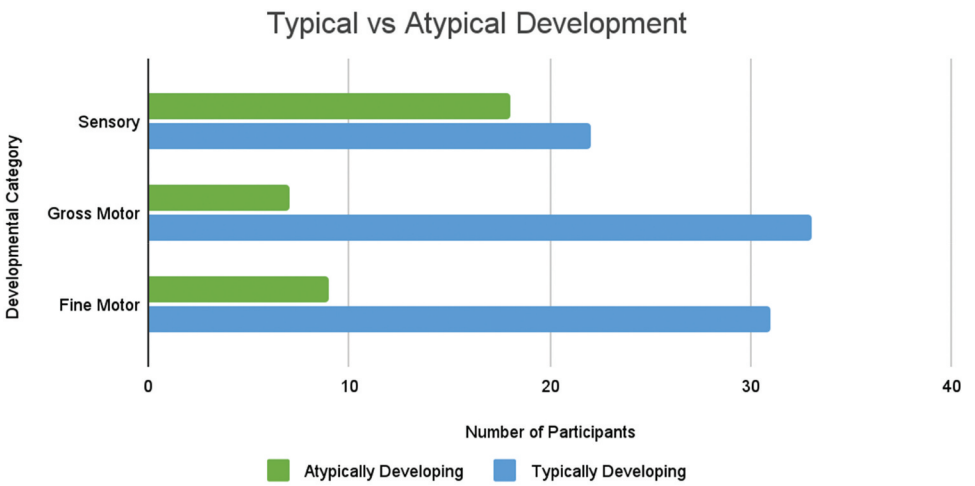

Figure 1. Typical vs atypical development patterns.

for physical development and adaptive behavior both at .72 which is a very large coefficient and provides convincing evidence for criterion-prediction validity; and 3) for construct validity, all 10 coefficients reported are statistically significant beyond the .0001 level, ranging from .42 to .75, which indicates that it is measuring what is purported to measure (Voress & Maddox, 2013).

Intervention Provided

All children with CG received intervention, and the authors intend to examine the effectiveness of interventions in future data analysis. Following pre-assessment, the Motor Milestones teleconference sessions were conducted regularly by an occupational therapist. Telehealth interventions consisted of the teach-model-coach-review approach (Roberts, Kaiser, Wolfe, Bryant, & Spidalieri, 2014). This structure guides the collaboration between the therapist and caregiver, laying the foundation for caregiver education. Session interventions were based on Beautiful Beginnings curriculum (Raikes & Whitmer, 2005) and Pathways developmental resources (Pathways, 2022). Beautiful Beginnings curriculum is designed for children from birth to 36 months incorporating developmentally appropriate skills and activities within the family's natural environment (Raikes & Whitmer, 2005). Treatment in the Motor Milestones portion of the study concluded at 15 months, then post assessments were completed at 30, 42, and 54 months of age.

Results

Percentages were calculated to determine the number of participants who had atypical motor and sensory scores within the infant, toddler, and child age groups. These descriptive statistics provide an overview of the sensory motor developmental profile of young children with CG. The following general developmental patterns emerged regarding participants with CG: 7/40 (17.5%) showed delay in gross motor development, 9/40 (22.5%) in fine motor development, and 18/40 (45%) in sensory processing

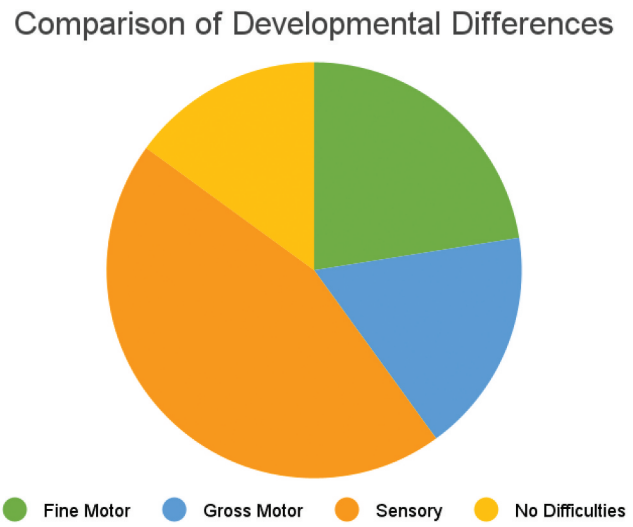

Figure 2. Comparison of delays among children with classic galactosemia.

(Figure 1). Of the participants who demonstrated atypical development, sensory processing differences accounted for almost half with fine motor and gross motor development, respectively (Figure 2).

Presence of Sensory Processing Patterns in Children with CG

Atypical development is defined as 1 standard deviation (SD) from the mean or in the "less than others" or "more than others" category of the Sensory Profile 2 (Dunn, 2014). Overall, seven out of forty (17.5%) participants scored "less than others" for sensory information, falling −1 SD below the mean. Eleven out of forty (27.5%) participants scored "more than others" falling +1 SD from the mean, and seven out of forty (17.5%) scored "much more than others" falling+2 SD from the mean. Of the 13 infants measured with the infant form, 0 showed differences in sensory processing. Of the 11 toddlers, nine (81.8%) showed differences in their sensory processing in the areas of sensitivity and seeking. No differences were identified in the areas of avoiding or registration. Of the 16 children, 9 (56.3%) showed differences in their sensory processing in the areas of seeking, avoiding, sensitivity, and registration to a lesser extent. Despite age, 18 out of 40 (45%) of the participants or slightly less than half showed differences in their sensory processing. When analyzing the overall sensory processing trends, it was found that participants ranked highest in sensitivity, seeking, and avoiding, respectively, with significantly less participants identified with registration (Figure 3). Analyzing each sensory system provides important insight into the sensory profile of children with CG. It highlighted a perplexing trend of the participants who began the study <6 months, and none were reported to have any differences in sensory processing. Interestingly, there was a large increase in sensory processing differences that surfaced starting around 7 months. Of the sensory processing differences, visual, auditory, oral, and movements were found to have the greatest differences among participants (Table 1).

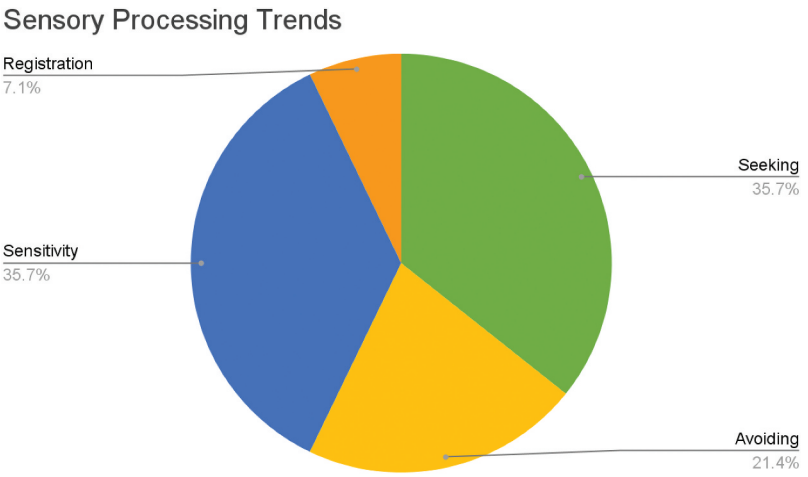

Figure 3. Sensory processing trends among children with classic galactosemia.

Table 1. The senses and individual differences.

The Senses & Individual Differences									
	Audi-tory	Visual	Touch	Movem-ent	Oral	Behavi-oral	Conduct	Social-Emotional	Attenti-onal
Infant	0	0	0	0	0	0			
Toddler	2	3	1	2	3	1			
Child	4	4	1	2	3		1	2	1

Presence of Motor Patterns in Children with CG

Atypical motor development was present for both cohorts, those who received the early motor intervention and those who received the speech/language intervention up to age 15 months. Within gross motor descriptive terms, 7 out of 40 (17.5%) were delayed in gross motor development, with all scores falling −1 SD from the mean. In fine motor development, there was a delay in 9 out of 40 (22.5%) from the mean. Seven of the participants fell −1 SD in fine motor development and two infants, −2 SD from the mean. When combining the fine and gross motor categories, the overall physical development yielded 22.5% of delays (Figure 1).

Presence of Adaptive Behavior Patterns in Children with CG

The adaptive behavior domain analyzes the independent, self-help functioning skills of children. After analysis, it was identified that 4 out of the 30 (13.3%) of participants have differences from the norm regarding these adaptive behaviors.

Discussion

To our knowledge, this is the first study to examine the sensory motor developmental trends of young children with CG. The proportion and distribution of atypical development in CG

infants is reported here in key areas relevant to multidisciplinary teams within Early Intervention (EI) and school-based therapy (SBT) settings, contributing to a paucity of developmental data in the CG population. Nearly (45%) of participants showed sensory processing differences in the sample with greater representation in the toddlers. This is a significant increase from 5–16% of all children in the general population, showing sensory processing differences (Ahn, Miller, Milberger, & Mcintosh, 2004; Ben-Sasson, Carter, & Briggs-Gowan, 2009). As such, this represents an important correlation that occupational therapy practitioners should be aware of when working with children with CG. Of the sensory processing trends, participants ranked high in sensory sensitivity, seeking, and avoiding with significantly less scoring in the registration category (Figure 3). This trend provides evidence regarding how a young child with CG may respond to sensory information. Those children may be more likely to be categorized as sensory seeker or sensory sensitive, which suggests an increase in behaviors indicative of increased engagement in sensory experiences or hyper-awareness of sensory input, respectively. Analyzing each sensory system of the participants found that visual, auditory, oral, and movement make up most of the sensory processing differences (Table 1). Interestingly, none of the infant participants scored out of a typical range in sensory processing, but, surprisingly, 82% of toddlers and 56% of children scored outside the typical range, indicating differences in sensory processing. Currently, the reason for this trend is unclear and the empirical data indicates potential areas of concern within sensory processing for children with CG.

The Galactosemia Network (GalNet) published an international guideline for the management of the metabolic condition including recommendations to screen for sensory processing difficulties and physical development with this population (Welling et al., 2017). In contrast to sensory processing, limited data has been reported regarding delayed motor development in children and adults at a rate of almost 27% (Rubio-Gozalbo et al., 2019). Data regarding the motor patterns show greater delay in fine motor compared to gross motor. The preliminary finding in this study identified fine motor discrepancies in 22.5% of children with CG,this rate is much higher than populations routinely screened for fine motor delays such as premature infants. Reported in a study by de Jong et al., moderate preterm infants' delay in fine motor skills was found to be 5.2% at 24 months of age (de Jong, Verhoeven, Lasham, Meijssen, & van Baar, 2015). This same study also shows similar rates of gross motor delay between moderate preterm infants (20.5%) as found in this preliminary study (17.5%) of children with CG (de Jong et al., 2015). These findings highlight the importance of including evaluation and screening of motor difficulties as a standard of practice when working with children with CG. From the preliminary data reported, developmental trends emerge for infants with CG, which should inform routine screening, evaluation, and intervention with this population.

Limitations and Future Directions

There are several limitations of the present study. First, the sample size was relatively small ($n = 40$). A larger sample size might reduce potential biases and increase generalizability. Second, there may be selection biases due to recruiting methods. Participants were recruited through websites, developmental physicians, online parent support groups, and word-of-mouth. Third, not all participants were administered the DAYC-2 adaptive subtest as this was a decision made later in the study process. Additionally, a literature review conducted

to examine adaptive behavior delays of other populations at or near 24 months resulted in zero studies. This did not allow the research team to analyze this data as a comparison with other populations. Additionally, a literature review conducted to examine adaptive behavior delays of other populations at or near 24 months resulted in zero studies. This did not allow the research team to analyze these data as a comparison with other populations. This excluded a handful of children who had already been evaluated skewing the data for this section. Fourth, this study examined post assessments at ages 30, 42, and 54 months. It is possible that other assessment intervals would be of interest or reveal insight into the data. Fifth, females were over-represented in the sample. Of the 40 participants in total, 27 were female and 13 were male. Sixth, The Sensory Profile assessment is a parent questionnaire on children's abilities. One possible explanation for the large jump in reported sensory difficulty is heightened awareness of these challenges through the BBC motor milestones treatment sessions. Other, possibly more objective, methods could be used to assess child abilities. Seventh, this study utilized standardized assessments that were not normed for Canadian and British participants.

As the clinical trial continues, the study will report on more extensive data sets. New participants continue to be added, generating data on a regular basis. Larger data sets will allow for comparison among a more complete extent of data. Future publications will build on these largely descriptive data, allowing for comparison among treatment groups, age, and gender. Longer-term outcomes will be evaluated allowing comparison of sensory motor development from entry into the study until age 4 years old. Additionally, future publications will include comparison of treated participants to untreated typically developing controls.

Professional Implications

Evidence from this study contributes important information regarding the sensory motor developmental profile of young children with CG. Preliminary evidence from the BBC study shows that proactive intervention beginning in infancy has beneficial effects on speech and language development (Peter et al., 2019, 2021). Analogous benefits for the developing motor and sensory systems should be investigated. This current and future research adds to a limited volume of evidence exploring the impact of CG on sensory motor and motor development patterns in young children with CG. This research lays the foundation for future studies with this population. These findings support improved awareness of the condition among occupational therapists, informing appropriate direction of assessment and intervention among occupational therapy practitioners working with these infants and children. Sensory processing screenings should be a routine part of care of this population to determine if it is a factor contributing to developmental differences. Application of this knowledge can inform the development and implementation of intervention programs to support optimal functioning and participation in daily life activities of children with CG.

Conclusion

This study adds preliminary findings necessary to identify the sensory motor developmental profile of young children with CG. Findings suggest the need for an increase in

research and evaluations regarding the impact of sensory differences on everyday life for a child with CG. As hypothesized, there are sensory-motor differences in the developmental profile of young children with CG. As the leading pediatric experts in evaluation and interpretation of developmental sensory motor characteristics, these findings bolster the need for occupational therapists to be integral members of the multidisciplinary teams for children with CG. In addition, knowledge of a child's physical development will be invaluable for caregivers and therapists creating interventions and activities to meet the individual needs of each child. There is a large research gap surrounding the implications of a CG diagnosis and sensory differences, however this paper brings awareness to the astounding preliminary data encompassing the sensory profile of children with CG. Occupational therapists are uniquely equipped to support infants and young children with CG navigate sensory experiences, adaptive strategies, support fine and gross motor development, and incorporate play to promote an increased functional independence in everyday occupations. As development progresses, having a strong foundation in these skills improves participation in daily activities into adulthood. These findings highlight the importance of including evaluation and screening of motor difficulties as a standard of practice when working with children with CG. Should the trends observed here become substantiated with future data, clinical implications for new intervention approaches will emerge, with the goal of improving early detection and intervention to prevent delay and solidifying occupational therapy's role in serving families impacted by CG.

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Disclosure statement

No potential conflict of interest was reported by the author(s).

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Authors' Contribution

The authors confirm contribution to the paper as follows: study conception and design: B. Peter, N. Potter, and M. VanDam; data collection: A. Monroe and C. Cupp; analysis and interpretation of results: A. Armstrong, A. Monroe, and C. Cupp; draft manuscript preparation: A. Armstrong, A. Monroe, C. Cupp, and B. Peter. All authors shaped the research and provided critical feedback to the manuscript.

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