

Research Article

Virtual Post-Intervention Speech and Language Assessment of Toddler and Preschool Participants in Babble Boot Camp

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ABSTRACT

Purpose: Babble Boot Camp (BBC) is a parent-implemented telepractice intervention for infants at risk for speech and language disorders. BBC uses a teach-model-coach-review approach, delivered through weekly 15-min virtual meetings with a speech-language pathologist. We discuss accommodations needed for successful virtual follow-up test administration and preliminary assessment outcomes for children with classic galactosemia (CG) and controls at age 2.5 years. Method: This clinical trial included 54 participants, 16 children with CG receiving BBC speech-language intervention from infancy, age 2 years, five children receiving sensorimotor intervention from infancy and changing to speechlanguage intervention at 15 months until 2 years of age, seven controls with CG, and 26 typically developing controls. The participants' language and articulation were assessed via telehealth at age 2.5 years. Results: The Preschool Language Scale-Fifth Edition (PLS-5) was successfully administered with specific parent instruction and manipulatives assembled from the child's home. The GFTA-3 was successfully administered to all but three children who did not complete this assessment due to limited expressive vocabularies. Referrals for continued speech therapy based on PLS-5 and GFTA-3 scores were made for 16% of children who received BBC intervention from infancy as compared to 40% and 57% of children who began BBC at 15 months of age or did not receive BBC intervention, respectively. Conclusions: With extended time and accommodations from the standardized administration guidelines, virtual assessment of speech and language was possible. However, given the inherent challenges of testing very young children virtually, inperson assessment is recommended, when possible, for outcome measurements.

Babble Boot Camp (BBC) is a proactive speech and language intervention designed for young children with a genetic or environmental risk for significant developmental delays (Finestack et al., 2022; Peter et al., 2019, 2021, 2022). The focus of BBC is to foster the earliest forms of spoken communication. All BBC intervention activities and routines were designed to be parent-implemented and incorporated into the child's daily routine in their natural environment. BBC is administered via telepractice by a speech-language pathologist (SLP) who trains parents using a teach-model-coach-review approach to recognize and reinforce the following six core communication components: (a) intentional eye gaze to support bonding, (b) responding to infant vocalizations to increase vocalization

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behaviors and to build dyadic interactions, (c) eliciting and reinforcing babble to increase babble complexity and to build associations between motor gesture and acoustic output, (d) labeling objects to expand the child's vocabulary, (e) modeling word productions to increase the child's phonemic inventory, and (f) recasting and expanding simple sentences to build syntactic complexity. The SLP, in collaboration with the parents, identifies speech and language skills that the child is capable of learning but has not yet mastered, consistent with operating within the zone of proximal development. Those skills are expected to develop relatively quickly, becoming the focus of the SLP's treatment plan (Smagorinsky, 2018).

BBC was designed to be delivered virtually and has the advantages of convenience, time efficiency, and reduced overall cost versus in-person therapy for parents of infants and toddlers (Behl et al., 2017; Campbell & Goldstein, 2022a; Thomas et al., 2017). Parents and children participate from home, eliminating preparation and commute time associated with outpatient models. One advantage of this flexibility is a reduction of cancellations. On average in the United States, patients do not show up for 18% of all outpatient appointments, with the no-show rates ranging from 5% to 55% (Berg et al., 2013). In contrast, the BBC families had only an 8% appointment noshow rate resulting in increased learning opportunities and decreased wasted provider time (Finestack et al., 2022). Virtual delivery saves families money by eliminating commuting and parking expenses. One additional advantage of the virtual delivery model for BBC is that parents are engaged throughout the therapy session and can participate even when the child is sleeping, thereby further reducing appointment cancellations and increasing attendance and retention.

The earlier a child is identified as having a risk for a developmental delay or disorder, the greater the likelihood that the child will benefit from targeted intervention (Bruder, 2010). Very early intervention can empower families and increase parenting skills and self-confidence, which in turn positively impacts a child's early learning and development. Early interventions have also been shown to be effective because they leverage brain plasticity and avoid the need to unlearn unintended behaviors (Blauw-Hospers & Hadders-Algra, 1999; Richter et al., 2017). Because BBC was designed to be delivered via telepractice, we were not limited by geographic area when deciding on our sample population; therefore, families who had children with rare conditions, residing in various locations in the United States and abroad, were eligible to participate in this study. We selected infants with classic galactosemia (CG) as the first population to trial BBC. The infants with CG were randomly assigned to one of two groups, Talk Time or Motor Milestones, at the time of enrollment between 2 and 6 months of age. Infants assigned to Talk Time began with weekly speech-language intervention while infants enrolled in Motor Milestones began with monthly motor intervention and transitioned to weekly speech-language intervention at 15 months of age. BBC intervention stopped for both groups when the children reached 24 months of age. CG is a rare recessive genetic disorder that occurs in one of approximately 30,000 live births in the United States, with a higher prevalence in individuals of Irish descent (Iwasawa et al., 2019). It is detected during the routine uniform newborn screening panel, typically conducted within the first 48-hr after birth. Individuals with CG have a near absence of an enzyme, galactose-1-phosphate uridyl transferase, which is needed to metabolize galactose, one of the sugars making up the lactose molecule and present in all mammalderived milk and milk products. After detection, affected infants are immediately placed on a lactose-restricted diet, which rescues the infant from serious kidney, liver, eye complications, and death, but not from the high probability of speech and language disorders (Fridovich-Keil & Berry, 2022). Early identification of a risk for a speech or language disorder and effective intervention to possibly minimize or prevent the disorder would be especially beneficial to the CG population as an estimated 60%-85% of affected children have long-term expressive language and speech disorders, including a 180-fold risk for childhood apraxia of speech (CAS; Overby & Highman, 2021; Potter et al., 2008; Timmers et al., 2011). Infants at risk for expressive language delays and CAS often show early indications of slower language development trajectories and delayed speech sound acquisition as compared to their typically developing (TD) peers, with many infants not meeting cooing and babbling milestones (Highman et al., 2012, 2013).

To evaluate the effectiveness of BBC, infants and toddlers are assessed in an ongoing longitudinal study with standardized indirect and direct assessments. The Preschool Language Scale–Fifth Edition (PLS-5; Zimmerman et al., 2011) and the Goldman-Fristoe Test of Articulation–Third Edition (GFTA-3; R. Goldman & Fristoe, 2017), assessments with sound psychometric properties, are directly administered to assess early speech and language abilities. Originally, the BBC intervention was designed to be delivered via telepractice, with follow-up assessments completed in person and conducted by local SLPs. The PLS-5 and GFTA-3 were selected because most early childhood SLPs are familiar with these assessments.

A challenge precipitated by the COVID-19 pandemic was the need to implement virtual assessments with toddlers. The publisher of the PLS-5 and GFTA-3 advised that professionals should use their clinical judgment to determine if an assessment via telepractice was appropriate for the examinee and situation (Pearson Corporation, 2021a, 2021b; Wright et al., 2020). They further stated that documentation of all considerations, procedures, and conclusions remained a professional responsibility, suggesting that remote administration could be conducted without serious violations of validity or an inability to interpret the test. In addition, the American Speech-Language-Hearing Association (ASHA) advised that standard evaluations may require modifications that may impact the interpretation of scores but does not rule out tele-administration of standardized tests, including the GFTA-3 and PLS-5 (ASHA, 2021; Freekmann et al., 2017).

The PLS-5 and the GFTA-3 are available in digital and print formats, but validity and reliability have not been formally and rigorously reported in the literature for remote or virtual administration. A concern with the virtual assessment of young children is that behaviors may interrupt administration and challenge interpretation. The GFTA-3 instructional manual indicates that the assessment is appropriate for administration in a comfortable, quiet, well-lit, home, clinical, or school environment with minimal distractions (R. Goldman & Fristoe, 2017). According to the GFTA-3 manual, the SLP may repeat items if the child is off task, is distracted, has missed the prompt, or if administration is interrupted. This flexibility to re-administer items makes the GFTA-3 optimal for remote administration. In one report in the literature, Campbell and Goldstein (2022b) administered the GFTA-3 to children ages 3-8 years, most diagnosed with speech sound disorders including CAS (Campbell & Goldstein, 2022b). They reported that there were one or more incidents that disrupted scoring on 30% of in-person and 50% of virtual assessments, but these incidents did not compromise accurate speech sound scoring and virtual and inperson administration vielded equivalent test results for individual sounds and standard scores. Thus, there is some successful precedent and discussion in the literature in support of remote administration of communication tests such as the GFTA-3.

The PLS-5 instructional manual states that the assessment is appropriate for administration in an area of the child's home with plenty of light and minimal distractions with extraneous stimuli removed from the testing area (Zimmerman et al., 2011). Children ages 1–3 years may be seated on the floor or at a table. The child may receive credit for passing specific test items if the caregiver provides specific examples even if the examiner did not observe the child demonstrating the behavior. Unlike the GFTA-3, in which all visual stimuli are presented on a screen, the PLS-5 alternates the stimuli among screen presentation, verbal instructions, and manipulatives; therefore, the PLS-5 manual states that the examiner should

develop competence with assessment via telepractice through activities such as practicing, studying, consulting with other professionals, and engaging in professional development. The PLS-5 has been used in other telepractice studies with young children, with one study explicitly using the PLS-5 and its subscales to compare telepractice and an in-person early intervention delivery (Behl et al., 2017). Another study examining remote administration of speech and language tests suggests that the PLS-5 should be adapted cautiously for remote administration because it is "designed to provide diagnostic information...[and] would not capture skill growth as well as studies designed for broader purposes" (Lund & Werfel, 2022, p. 401). Thus, there is some precedent for remote administration of the PLS-5, but it is an emerging tool in this modality and future work is needed to better understand it.

The full assessment protocol for BBC includes direct and indirect approaches. Each has its own advantages. Indirect approaches such as surveys or parent questionnaires typically require less time and effort to complete. Direct approaches require the commitment of the family, child, and assessor, typically over a longer time period and often multiple sessions (Ollington, 2016). We report on the procedures and outcomes of direct assessment of children in BBC at ages 2-2.5 years of age. Previous publications have demonstrated that BBC intervention can be successfully implemented with high parent satisfaction and shows early indications of boosting babble and early speech production (Finestack et al., 2022; Peter et al., 2019, 2021, 2022). In this report, we examine early outcomes, along with the advantages and challenges, of assessing receptive, expressive language, and articulation skills of children in BBC at 2.5 years of age via telepractice. Specifically, we asked the following research questions.

- 1. Can the PLS-5 or GFTA-3 be successfully administered virtually to assess toddlers' speech and language skills? If yes, what accommodations facilitate successful administration?
- 2. Does virtual test administration duration differ from in-person test administration duration?
- 3. Do the follow-up assessments show treatment effects for the children in the BBC Talk Time treatment group compared to treatment initiated at 15 months for the BBC Motor Milestones treatment group or the children in the no treatment control groups?

Method

Parents provided informed consent to participate in this study for themselves and provided written permission for their child's participation. The study was reviewed and approved by the institutional review boards at Arizona State University (STUDY00004969) and Washington State University (13099–014, 19422–001) and is registered at ClinicalTrials.gov under Beate Peter and at Open Science Framework under https://osf.io/yzht4/.

Participants

The study included 28 children with CG and 26 TD children. To be eligible for the study, the children with CG had a confirmed genetic neonatal diagnosis with no evidence of other chromosomal, sensory, or medical conditions. Children with CG were recruited through referrals by health care providers, via social media or online announcements on the Galactosemia Foundation website (https://galactosemia.org). The families of the children with CG were enrolled in this study as soon as possible following referral or initial contact and always within a month following their referral.

All BBC families needed to have at least one parent who completed high school or earned their general education diploma. Most parents (69%) had earned a bachelor's or graduate degree. Participating families spoke English as their primary language and lived in the United States (n =29) or the United Kingdom (n = 3). Internet access was required, and all families already had computers and Internet access. Most families identified their child as White, not Hispanic (n = 31). One family identified their child as Black. The limited diversity in our participant pool reflects the genetic risk for CG being concentrated in people of European, predominantly Irish, decent (Iwasawa et al., 2019).

BBC TD controls were volunteers known to the research team or recruited by word of mouth or social media. They were a sample of convenience and were included as age-matched controls because only a few children in the BBC TD control group (n = 4) had reached 2.5 years of age at the time of this report. The TD controls were from the HomeBank (VanDam et al., 2016)

and the Cougar Corpus repositories (VanDam, 2018), together referred to as HomeBank TD controls in this report. The data for this study were collected from the HomeBank repository because they represented comparable demographic characteristics and included GFTA-3 for all the children. None of the TD children received intervention. The HomeBank TD children had at least one parent who completed high school or equivalent, all spoke English as their primary language, and all lived in the United States. All identified their children as White, and one identified as Hispanic. All HomeBank TD controls reported no disabilities or developmental concerns.

Demographic and group details are included in Table 1. CG Infants were randomly assigned to engage in BBC at two different time points. The BBC Talk Time group included 16 children with CG who received weekly BBC speech-language intervention from the time of enrollment at ages 2-6 months until 24 months of age. The BBC Motor Milestones group included five children with CG who received biweekly sensorimotor/occupational therapy intervention from the time of enrollment, at ages 2-6 months, through age 14 months and then discontinued sensorimotor therapy intervention and began weekly BBC speech-language intervention from 15 months until 24 months of age. The Motor Milestones group was included as a control group with less frequent intervention during the sensorimotor/occupational therapy intervention period to examine if the predicted long-term positive outcomes were related to the provision of parent-implemented intervention in general during infancy or to the specific BBC early speech-language intervention. The BBC CG control group included a total of seven children. One child enrolled in the study between 2 and 6 months of age and was randomized into a control group. Six children enrolled as toddlers and did not receive intervention. The families of the children with CG enrolled as toddlers had either not heard about BBC before their child reached 6 months of age or the added demands of the pandemic prevented them from responding to the study announcements. The BBC

Table 1. Demographic and group details of participants.

Group	Males	Females	Total <i>n</i>	Diagnosis
BBC Talk Time	8	8	16	CG
BBC Motor Milestones	1	4	5	CG
BBC CG controls	2	5	7	CG
BBC TD controls	1	3	4	TD
HomeBank TD controls	11	11	22	TD
Total participants with CG	11	17	28	CG
Total TD participants	12	14	26	TD
Total participants	23	31	54	Mixed

Note. Talk Time and Motor Milestones are two different treatment arms of Babble Boot Camp. HomeBank is an online, public corpus including TD children used here as controls. BBC = Babble Boot Camp; CG = classic galactosemia; TD = typically developing.

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TD controls were four children who did not have CG or any other known diagnosis negatively affecting development and did not receive BBC intervention. The Home-Bank TD controls were children ages 22–34 months, mean age of 28.5 months, did not have intervention, and had been assessed with the GFTA-3, but not the PLS-5.

The data reported here are part of an ongoing BBC clinical trial. Participants in the BBC pilot study were infants assigned to the Talk Time group, accounting for the larger number of children in this group who have reached the age of at least 2.5 years compared to the other groups. While new participant enrollment for the CG population has been completed, most of the children in Motor Milestones and the controls have not yet reached age 2.5 years and some are still under 1 year of age.

Measures

The GFTA-3 is an articulation test for ages 2:0-21:11 (years;months); the Sounds-in-Words subtest assesses initial medial and final sounds for 23 consonant and 15 consonant blends in 60 target words. Children name or attempt to name all 60 target words. The PLS-5 is a comprehensive developmental language assessment for ages birth to 7;11. The Auditory Comprehension subtest (PLS-5 AC) assesses receptive language skills, whereas the Expressive Communication subtest (PLS-5 EC) assesses expressive language skills. The PLS-5 AC includes 65 numbered items, and the PLS-5 EC includes 67 numbered items. The starting item is based on the child's chronological age. A basal is achieved when a child responds correctly to three consecutive items, and a ceiling is achieved when a child misses six consecutive items. Time to complete assessments was computed post hoc from recordings and session notes. For the PLS-5, only the number of hour-long sessions was recorded due to time-out interruptions reducing the overall ability to accurately record testing time including re-administering earlier correctly answered items to engage the child at the beginning of a new session, short breaks for the child to eat or drink, toileting and diapering breaks, the child leaving the testing area to retrieve a different preferred item, and sibling interruptions. GFTA-3 testing time was recorded in minutes.

Procedure

Children's speech and language were assessed, using the PLS-5 and GFTA-3, at age 2.5 years, 6-months after they had completed BBC intervention. These two assessments were administered in connection with a battery of indirect standardized tests, not further reported here. Early in the pandemic, our research team tried virtually administering these assessments to two children at 24-months of age. The children had difficulty staying focused on the screen in their home environment and crawled onto the table or left the testing area. Following the assessment attempt with 24-month-olds, the BBC team made the decision to move the assessment ages to 2.5, 3.5, and 4.5 years to increase success rates. Three 1-hr virtual assessment sessions were scheduled on separate days within 2-weeks for each child. The GFTA-3 and items from the PLS-5 AC were completed during Session 1. The remaining items from the PLS-5 AC and all of PLS-EC were completed during Session 2 and Session 3, if needed. Assessment sessions were discontinued when the child exhibited fatigue or could not be redirected to the assessment. Children were allowed short breaks as needed. Assessments were conducted and recorded over Zoom with pictures presented electronically over a shared screen. Two examiners were present on Zoom for > 80% of the assessment sessions. A single examiner was present for all other assessment sessions. When two examiners were present, one examiner shared their screen and provided the question, model, or instructions to the parent or child, per the standardized administration guidelines in the assessment manuals. Both examiners independently recorded the child's answers. In addition, the second examiner observed the child and wrote notes regarding the child's responses. The assessment team was blinded to the child's diagnosis and treatment group membership.

PLS-5

Prior to the first assessment session, parents were e-mailed a list of objects to assemble to substitute for the manipulatives in the PLS-5 test kit. All parents had the required items in their home and did not need to purchase additional materials. Parents were actively involved in keeping the child's attention focused on the assessment and manipulating objects for the PLS-5. The PLS-5 manual states that items requiring manipulatives cannot be administered remotely unless a specially trained facilitator is at the remote site (Zimmerman et al., 2011). If such a facilitator is well trained and in a professional role (i.e., a professional facilitator), he or she can be provided with training to administer test items that have manipulatives specific to PLS-5, as well as adjust audiovisual equipment. In times when physical distancing is necessary (such as the COVID-19 pandemic), using a professional facilitator may not be safe or feasible. If testing must occur under these conditions, it is possible that the examinee may participate with a caregiver as an on-site facilitator to assist the child during the test session. For the PLS-5, parents were given specific instructions for test items with manipulatives immediately before asking the child to complete the task (e.g., "Scatter eight crayons on the table"). When specific instructions would compromise the child's response,

instructions were spelled (e.g., "Put the spoon U-N-D-E-R the box"). The sequence of items on the PLS-5 alternates between the child viewing pictures in the manual or on the screen and manipulating objects. The manual's instructions for item administration state that the items may be administered out of sequence to maximize the child's performance. We administered groups of test items requiring manipulating objects and groups of items requiring attention to the screen to minimize transitions for children and their parents who were facilitating the assessment. Parents verbally indicated "R" for right and "W" for wrong for responses that were not visible to the examiner.

Parents may be concerned when their child misses the six consecutive items needed to reach a test ceiling on the PLS-5. The PLS-5 manual instructs that the examiner should explain to a parent or caregiver who is present during the assessment that some of the items will exceed the child's abilities. To minimize parental distress, we explained that we would be testing beyond the child's abilities and stated when the child was at the starting point for ages 4, 5, 6, or 7 years. Since the PLS-5 administration was spread out over two to three sessions with breaks as needed, often the assessment was discontinued mid-subtest and resumed on the next scheduled day. The examiners intentionally used repetition of an earlier correctly answered item to increase the child's initial success to start the session. The PLS-5 was scored online when two examiners were present or with video review as needed when only one examiner was present.

GFTA-3

The GFTA-3 test manual directions suggest using the question, "What is this?" to elicit target responses for most items (R. Goldman & Fristoe, 2017). It is acceptable for the examiner to vary the stimulus question, but the item's name should not be included in the prompt. Individuals may also name the item without a prompt. If the individual cannot name the item, the examiner may provide the item's name followed by an intervening sentence without the item's name. The manual does not state if, or what, modifications were needed to elicit the targets with toddlers. In this study, the GFTA-3 was administered per standardized directions by asking the child, "What is this?" or waiting for the child to name the target. If the child was unable to name the target the examiner stated, "This is a _____. What is it?" If the child did not name the target or the response was incorrect, the target was elicited in imitation. The GFTA-3 was scored online by both examiners and postadministration by consensus with an experienced SLP/researcher and a graduate student. The SLP/researcher (N.L.P., first author) had clinical and research expertise in galactosemia and the assessment of severe speech sound disorders, including CAS. Following each GFTA-3 assessment, the examiners completed an item-by-item comparison. All questionable phoneme productions and any scoring discrepancies between examiners were resolved by consensus after watching and listening to the recording of each target item a maximum of 3 times. When only one examiner was present, the same examiner rescored the entire assessment after watching and listening to the recording of each target a maximum of 3 times. Any item that was in question was video-reviewed and rescored by the consensus of two examiners. A child's production was considered correct if there was > 50% adultlike accuracy on manner, place, and voicing. We used a consensus process, listening to the recording, independently judging manner, place, and voicing, followed by discussion among examiners, to resolve discrepancies for two reasons. First, one of the examiners was a graduate student who was learning to score the GFTA-3; and second, it is difficult to judge 2.5-year-olds' phoneme production as correct and incorrect.

Standard Scores

For this study, "within normal limits" was defined as a standard score between 85 and 115 on the PLS-5 and GFTA-3. Both assessments are normed to a population mean of 100 and a standard deviation of 15. A standard score of 85 corresponded to a PLS-5 AC raw score of 29, PLS-5 EC raw score of 28, and GFTA-5 raw score of 85– 88. Following the completion of all the assessments, the examiners met and referred children with standard scores less than 85 on the PLS-5 or GFTA-3 for further inperson speech-language evaluation by a local SLP. Based on the number and type of errors, the examiners also recorded whether CAS could be reasonably ruled out.

Analyses

Demographic and group variables were entered into models as independent variables, and GFTA-3 standard scores, PLS-5 standard scores, and testing time were treated as dependent variables in separate models. The data did not meet the assumptions of the parametric model, for example, the assumptions of a reasonably large number of observations was not maintained and, by visual inspection of the histogram, the data did not clearly constitute a normal distribution. We compared pairwise groups for difference using the nonparametric Wilcoxon-Mann-Whitney test. Alpha threshold was set to .05. To estimate effect sizes, all pairwise comparisons were considered using Hedge's g. Hedge's test is appropriate here because it gives relative weighting to the distributions under consideration according to sample size (Grissom & Kim, 2005). Because of this attention to (dissimilar) sample sizes, Hedge's test is

Table 2. Standard scores and standard deviations for the GFTA-3, PLS-5 AC, and PLS-5 EC for all groups.

Assessment (standard score/standard deviation)	BBC Talk Time	BBC Motor Milestones	BBC CG controls	BBC TD controls	HomeBank TD controls
GFTA-3	105.6/15.1	96.2/19.5	81.7/21.5	106.7/27.2	98.5/11.7
PLS-5 AC	103.6/11.9	103.8/10.6	93.8/10.4	113.7/18.7	
PLS-5 EC	100.3/9.8	102.8/16.7	90.4/14.9	103.7/21.4	

Note. GFTA-3 = Goldman-Fristoe Test of Articulation–Third Edition; PLS-5 AC = Auditory Comprehension subtest of the Preschool Language Scale–Fifth Edition; PLS-5 EC = Expressive Communication subtest of the Preschool Language Scale–Fifth Edition; BBC = Babble Boot Camp; CG = classic galactosemia; TD = typically developing.

suitable for the present data. The Hedge's g statistic is interpreted in terms of standard deviations difference-scores between the samples.

Results

All the children in BBC at age 2.5 years were able to complete the PLS-5 AC and EC subtests by achieving basal and ceiling scores. In the sample reported here, the PLS-5 standard scores did not differ between any groups based on pairwise comparisons (all p values > .1); however, it is noted that the statistical tests have a modest number of participants in the Motor Milestones and control groups. The preliminary results reported here show that the children in the BBC Talk Time and BBC Motor Milestones intervention groups had PLS-5 AC mean standard scores of 103, indicating typical language development, which is above the scores for the BBC CG controls but below the BBC TD controls (see Table 2). PLS-5 EC standard scores were comparable across the BBC Talk Time, BBC Motor Milestones, and BBC TD control groups but lower in the BBC CG control group. Summary statistics including means, standard deviations, test statistics, observed probability values, and effects sizes represented by Hedge's g are shown in Tables 2–5. Overall, three children had PLS AC standard scores above 115 (two from Talk Time, one from the BBC TD control group) and two children (both from the BBC CG control group) had standard scores below 85. The remaining 27 children were within normal limits of 85-115. On the PLS-5 EC subtest, three children had standard scores above 115 (one from Talk Time, one from Motor Milestones, one from the BBC TD control group) and five had standard scores below 85 (one from Talk Time, one from Motor Milestones, two from BBC CG control group, one from the BBC TD control group). The remaining 24 children were within normal limits of 85–115.

None of the BBC participants were able to spontaneously name all the GFTA-3 targets (e.g., "vegetable," "pajamas," "crown") and required a model followed by the question, "What is this?" or direct imitation of the examiner's model. All children except three were able to complete the GFTA-3. The three children (all in the CG control group) who attempted the GFTA-3 were unable to complete it due to limited expressive vocabularies (e.g., [e.g., bAbA] for most responses), simplified syllables (e.g., [po] for "apple"), and single phonemes (e.g., [p] for "pig") to approximate the targets. For the GFTA-3 comparisons, the BBC CG control group had lower standard scores than the BBC TD control group (U = 2.34, p = .019). No other comparisons reached statistical significance. The preliminary results reported here show a trend toward similar performance on the GFTA-3 for the BBC Talk Time group and BBC TD control groups with slightly better articulation skills than the BBC Motor Milestones and significantly better than the BBC CG control group. Overall, nine children had GFTA-3 standard scores above 115 (four from Talk Time, one from Motor Milestones, two from the CG control group, two from the HomeBank TD control group), and 10 children had standard scores below 85 (one from Talk Time, two from Motor Milestones, four from the CG control group, one from the BBC TD control group, two from the HomeBank TD control

Table 3. Number of children with a Goldman-Fristoe Test of Articulation-Third Edition (GFTA-3) raw score of > 85 (raw score = number of errors).

Group	Male	Female	n/total	Diagnosis
BBC Talk Time	1/8 (13%)	1/8 (13%)	2/16 (13%)	CG
BBC Motor Milestones	1/1 (100%)	1/4 (25%)	2/5 (40%)	CG
CG no BBC intervention	2/2 (100%)	2/5 (40%)	4/7 (57%)	CG
BBC TD controls	0/1 (0%)	1/3 (33%)	1/4 (25%)	TD

Note. Raw score of 85 corresponds to a standard score of < 85 or -1 *SD* on the GFTA-3. BBC = Babble Boot Camp; CG = classic galactosemia; TD = typically developing.

Mann–Whitney U (p/z value)	Group	BBC Talk Time	BBC Motor Milestones	BBC CG controls	BBC TD & HomeBank TD controls
GFTA-3	MM	.26/1.11			
	CG ctr	.019/2.34	.32/0.98		
	TD ctr	.70/-0.37	.54/-0.61	.10/-1.62	
	TDHB	.066/1.83	.66/-0.43	.07/-1.76	.33/0.96
PLS-5 AC	MM	.96/-0.04			
	CG ctr	.13/1.50	.22/1.22		
	TD ctr	.29/-1.04	.45/-0.75	.08/-1.70	
PLS-5 EC	MM	.61/-0.49			
	CG ctr	.15/1.40	.28/1.06		
	TD ctr	.96/-0.04	.99/0.01	.39/-0.85	

Table 4. Observed probability and Mann–Whitney U scores (given as z scores) for each pairwise comparison.

Note. BBC = Babble Boot Camp; CG = classic galactosemia; TD = typically developing; GFTA-3 = Goldman-Fristoe Test of Articulation–Third Edition; MM = Motor Milestones; ctr = control; TDHB = typically developing HomeBank; PLS-5 AC = Auditory Comprehension subtest of the Preschool Language Scale–Fifth Edition; PLS-5 EC = Expressive Communication subtest of the Preschool Language Scale–Fifth Edition.

group). The remaining 35 children were within normal limits of 85–115. Summary descriptive statistics are given for all tests and all groups in Figure 1 and Table 2.

About half of the children (52%) completed the PLS-5 and GFTA-3 in two 1-hr assessment sessions and the rest required three 1-hr sessions. The PLS-5 manual states that the assessment takes 45–60 min to administer. Virtual administration time was longer but could not be accurately calculated as there were interruptions for multiple reasons, as discussed in the procedure section, and most children required multiple breaks. The GFTA-3 manual states that the assessment takes 5–15 min to administer. Virtual administration time for this assessment at age 2.5 years ranged from 9 to 33 min with a mean of 19 min.

Following the assessments, the SLP/researcher and one graduate student on the assessment team discussed the child's performance and follow-up recommendations. We determined that CAS or other severe speech disorders could be not ruled out if the child showed early signs of speech delay/disorder with decreased intelligibility, GFTA-3 standard scores below 85, and errors characterized by omissions, substitutions, vowel errors, consonant voicing errors, and distortions. CAS could not be ruled out, and further speech-language evaluation and intervention were recommended for a total of nine children, 57% (n = 4) of the children in the BBC CG control group, 40% (n = 2) of the children in the BBC Motor Milestones, 25% (n = 1) in the BBC TD control group, and 13% (n = 2) of the children in the BBC Talk Time group. Of the nine children referred for further in-person speech-language follow-up, two had PLS-5 AC standard scores and four had PLS-5 EC standard scores below 85. No children were referred for in-person speech-language based on their PLS-5 standard scores alone. Only one other child had a PLS-5 EC standard score slightly below 85, but this child showed

Effect size, Hedge's <i>g</i>	Group	BBC Talk Time	BBC Motor Milestones	BBC CG controls	BBC TD & HomeBank TD controls
GFTA-3	MM	0.622			
	CG ctr	1.391	0.699		
	TD ctr	0.062	0.454	1.061	
	TDHB	0.536	0.173	1.161	0.562
PLS-5 AC	MM	0.017			
	CG ctr	0.852	0.954		
	TD ctr	0.76	0.676	1.448	
PLS-5 EC	MM	0.215			
	CG ctr	0.861	0.792		
	TD ctr	0.031	0.047	0.767	

Table 5. Effect sizes represented by Hedge's g for each pairwise comparison.

Note. BBC = Babble Boot Camp; CG = classic galactosemia; TD = typically developing; GFTA-3 = Goldman-Fristoe Test of Articulation–Third Edition; MM = Motor Milestones; ctr = control; TDHB = typically developing HomeBank; PLS-5 AC = Auditory Comprehension subtest of the Preschool Language Scale–Fifth Edition; PLS-5 EC = Expressive Communication subtest of the Preschool Language Scale–Fifth Edition.

Figure 1. Means (triangles) and standard deviations (vertical lines) across groups for the GFTA-3 (articulation), PLS-5 AC (receptive language), PLS-5 EC (expressive language) standard scores. GFTA-3 = Goldman-Fristoe Test of Articulation–Third Edition ; PLS-5 AC = Auditory Comprehension subtest of the Preschool Language Scale–Fifth Edition; PLS-5 EC = Expressive Communication subtest of the Preschool Language Scale–Fifth Edition; TT = Talk Time (black); MM = Motor Milestones (green); CG = BBC Classic Galactosemia untreated controls (aqua); TD = typically developing controls (red); HB = HomeBank typically developing controls (blue); BBC = Babble Boot Camp.



signs of being distracted during this subtest and had PLS-5 AC and GFTA-3 standard scores within normal limits so was not referred for follow-up at the time of testing. This child will be referred if standard scores are less than 85 when assessed at age 3.5 years.

Discussion

This study examined the follow-up speech and language assessments of children in BBC Talk Time and Motor Milestones at age 2.5;6 after their BBC intervention ended, and controls who did not receive BBC intervention. We aimed to describe what accommodations were needed for the successful administration of the PLS-5 and GFTA-3 and whether the follow-up assessments showed treatment effects.

Feasibility of Virtual Assessments

BBC intervention is well suited for telepractice administration with parents reporting that they valued the information and support, the intervention was welldesigned, and their child benefited from participating in BBC (Finestack et al., 2022). The virtual direct assessment of 2.5-year-olds with standardized tests was challenging and less well suited than the intervention, but still possible. The original plan for the BBC clinical trial was for parent-coaching to be virtual with direct follow-up assessment by a local SLP at participant ages 2, 3, and 4 years using the PLS-5 and GFTA-3. The publisher of the PLS-5 and GFTA-3 advised that professionals should use their clinical judgment to determine if an assessment via telepractice was appropriate for the examinee and situation, but due to the Covid-19 pandemic, all assessments moved to a virtual administration (ASHA, 2021; Pearson Corporation, 2021a, 2021b; Wright et al., 2020). The PLS-5 manual states that the examiner should develop competence with assessment via telepractice through activities such as practicing, studying, consulting with other professionals, and engaging in professional development. The SLP/researcher had more than 15 years of experience using telepractice for assessment and intervention prior to the pandemic. These factors taken together are all consistent with the letter and the spirit of test administration and interpretation.

Early in the pandemic when the surface transmission risk was unknown, we asked parents to assemble a kit of manipulatives for the PLS-5 from a list sent in advance rather than mail a set of manipulatives that carried an unknown risk of disease transmission (E. Goldman, 2020; Greenhalgh et al., 2021; Zimmerman et al., 2011). An unintended positive consequence of using test manipulatives from the child's environment was that the familiar items acted as natural reinforcers. Natural reinforcers are familiar items that have high availability within the child's natural environment and serve to facilitate a child's successful performance (Los Horcones, 1992). Parents reported that they chose the child's preferred items to use during test administration. Children engaged quickly with their favorite stuffed animal, substituted for the plastic purple bear from the PLS-5 test kit, when asked to share their crayons with their stuffed animal or give it a drink. The greatest challenge with using parents as facilitators, rather than the recommended professional facilitator for the assessments, was that all parents want their child to succeed so watching their child miss six consecutive items to reach the PLS-5 ceiling was stressful for parents (Zimmerman et al., 2011). Parents were noticeably more at ease serving as the test facilitator when the examiners stated, "We are now at the starting point for age __." At times, parents started to restate the PLS-5 questions or instructions when their child did not appear to understand. In these instances, parents were gently reminded that they may provide a repetition but may not change the wording.

Assessment Time

The parents' primary responsibility during the administration of the GFTA-3 was to keep the child engaged and redirect their attention to the screen as needed. Some parents asked if they could use a food item to reward their child for attending and participating during the assessment. We requested that parents withhold food items during the GFTA-3 administration and, while

not encouraged, we allowed food reinforcers during the PLS-5 administration. Most 2.5-year-olds have limited attention spans and do not typically return to a previous location or return their attention to a previous activity following a pause or a break. Offering a snack in the testing area seemed to encourage the children to remain in the assessment location (Sarid & Breznitz, 1997).

The mean administration time of 19 min slightly exceeded the publisher's 15-min maximum estimated time required to administer the GFTA-3 in person as stated in the manual with most children completing the GFTA-3 without needing a break (R. Goldman & Fristoe, 2017). Considering that the published administration time was inclusive of ages 2-21 years and that in-person administration time for the GFTA-3 for children ages 2-3 years was not specifically stated, it would be reasonable to assume that young children would likely require more time compared to the administration time with older children. Parents appeared comfortable during the GFTA-3 assessment and often were surprised by their child's spontaneous naming of items they did not predict the child could name. The examiners chose to discontinue the GFTA-3 assessment for the three children with limited expressive vocabulary and lexical inventories when the parent or child showed signs of frustration and distress.

Treatment Effects

The early results of the direct follow-up assessments indicate that BBC parent-implemented very early intervention, whether speech-language or motor focused, appeared to positively impact language development. Nearly all the children in the BBC Talk Time group, who received weekly speech-language intervention from infancy to age 2 years, and the BBC Motor Milestones group, who received biweekly motor intervention from infancy through age 14 months and then weekly speech-language intervention until age 2 years demonstrated typical language development at 2.5 years of age while 29% of the CG control group, who received no intervention, had receptive and expressive language delays. Speech-language disorders are estimated to affect 50%-60% of the children with CG who have had standard care, with CAS affecting about 24% of the children with speech disorders (Shriberg et al., 2011). Most children with CG have expressive language disorders with normal or near-normal receptive language (Potter et al., 2008). Expressive language delays were less prevalent than expected in our sample. Using the PLS-5 standard score of less than 85, only two children, both in the CG control group, had receptive and expressive language delays, and an additional three children had only expressive language delays (one each from Talk Time, Motor Milestones, and BBC TD control group). Across all the CG groups, language delays were less prevalent than we expected based on other studies examining language development in CG. One possible explanation is that, as discussed by other assessment teams, the PLS-5 is not sensitive for detecting subtle expressive language delays and disorders at age 2.5 years (https://community.asha.org/viewdocument/ pls-5-validity-study?CommunityKey=f64f6543-3ba0-445e-bbe7a14586106c59&tab=librarydocuments). The BBC study follows children until 4.5 years of age, and we predict that the children with CG in the control group will have a higher prevalence of language delays as more complex language skills are expected to emerge.

Our finding that children who received BBC Talk Time intervention from infancy to 2 years of age had fewer speech errors when assessed at age 2.5 years than infants with no intervention supports the premise that BBC speech-language intervention reduces the risk of later speech delays. By intervening during early infancy, babies were exposed to frequent and repetitive correct speech sound models and given many opportunities to produce phonemes and phoneme combinations within their zone of proximal development. We propose that targeting the specific precursors of speech, a baby's first words, and later phrases during the rapid neuroplastic changes that occur in children under the age of 2 years minimizes acquiring and habituating errored sound patterns (Natu et al., 2021). Another possible explanation for the higher performance in the Talk Time group, compared to the Motor Milestones group, is that the intense focus on babble quantity and complexity during the prespeech phase served to solidify motor-sound associations that can later support speech sound production. It is reasonable to expect that with standard care, more than 50% of the children with CG in each BBC group would show indications of a significant speech delay or disorder and would have a high risk of CAS. However, there are no universally accepted standards for prevalence or type of risk factors that lead to a later CAS diagnosis in children under 3 years of age (Overby & Highman, 2021). In older children, one tool referred to as the Mayo 10 is the observation of at least four of the 10 signs of CAS in three different speaking situations (Shriberg et al., 2011, 2017). The Mayo 10 signs require that the child can produce some conversation with novel multiword utterances, which is a developing skill at age 2.5 years. We were able to observe the children's performance in this study on two different standardized assessments. Following the assessments, the SLP/researcher and one graduate student on the assessment team discussed the child's performance and followup recommendations. The children who were referred had more than 85 errors on the GFTA-3, and multiple vowel errors and voicing errors on consonants, which are the first two signs on the Mayo 10 list, in addition to

consistent and inconsistent substitutions, omissions, and some distortions. These observations are not meant to be a short list of CAS diagnostic criteria but rather a means to compare across the different groups of children in BBC. Young children have heterogeneous speech development; thus, a delay at 2.5 years is not necessarily indicative of a persistent speech disorder. However, as supported by the CAS literature, early speech development of children later diagnosed with CAS differs from that of TD children in quantity and quality (Overby & Highman, 2021).

ASHA advises that assessment scores may differ across in-person and online administration (ASHA, 2021). Researchers (Campbell & Goldstein, 2022b) examining virtual versus in-person multiple examiner administration of the GFTA-3, with 39 children, ages 3-8 years with and without speech sound disorders, reported that the different test environments yielded equivalent standard scores. An earlier telepractice intervention study demonstrated that the PLS-5 was an effective outcome measure to differentiate between groups of toddlers receiving in-person or telepractice speech-language intervention, with the telepractice intervention group scoring higher than the in-person intervention group (Behl et al., 2017). The authors did not state whether the PLS-5 was administered in person or via telepractice for either group. While data comparing inperson versus telepractice performance on standardized assessment with children under age 3 years is not currently available, we confidently report standard scores here as a comparison among children and groups in this study since the same BBC team administered and scored the assessments via telepractice. Following the assessments, parents were sent summary reports with raw scores, standard scores, confidence intervals, and percentiles with the following caveat statement, "Assessments were not developed for online administration, so standard scores are estimates of performance only."

Limitations

The study is still in progress, so the results are preliminary. We were fortunate to have a telepractice study underway when the pandemic hit, but had we had an inkling that the clinical world would go on pause, we would have tested in-person and online administration of follow-up assessments prior to needing them. There is little diversity in our participant pool, which reflects the strong Irish risk factor for CG, but this may limit direct application to other populations. The HomeBank Control group was a sample of convenience that was comparable on age and GFTA scores, but that sample was not controlled in the same systematic ways as the large, more cohesive data from the CG families and other controls in the larger BBC study.

Conclusions

Overby and Highman (2021) made a plea for research to address the great unmet need for treatment studies focused on the infant and toddler population, especially children at risk for CAS. They challenged researchers to develop an effective, impactful, specific intervention to address our youngest clients' unique needs, attentional focus, memory, and learning style. BBC is an answer to this great unmet need with a practical parent-implemented approach for at-risk infants that can be administered by SLPs via telepractice in 20-min weekly sessions. Without intervention, we would expect that more than 50% of the children with CG would demonstrate delays in language and articulation development. Our early positive effect sizes show that at 2.5 years of age the children who received BBC intervention as infants until age 2 years were performing as well in receptive and expressive language and articulation at the phoneme in words level as the TD controls. In addition to showing early effectiveness, BBC intervention had high parent involvement and satisfaction.

We demonstrated that it is possible to successfully administer standardized assessments with parents as the test facilitators via telepractice with children as young as 2.5 years of age. In addition to access to a computer and Internet service, successful administration requires advance preparation to assemble substitute physical stimuli and multiple scheduled sessions. Although feasible, given the inherent challenges of testing very young children virtually, in-person assessment is recommended, when possible, for outcome measurements.

Author Contributions

Nancy L. Potter: Conceptualization (Equal), Investigation (Lead), Writing - original draft (Equal), Writing review & editing (Equal). Mark VanDam: Conceptualization (Equal), Methodology (Lead), Writing - original draft (Equal), Writing - review & editing (Equal). Laurel Bruce: Writing - review & editing (Supporting). Jenny Davis: Conceptualization (Equal), Investigation (Equal), Writing review & editing (Supporting). Linda Eng: Project administration (Equal), Writing - review & editing (Supporting). Lizbeth H. Finestack: Conceptualization (Equal), Methodology (Equal), Writing - review & editing (Equal). Victoria Heinlen: Investigation (Equal), Writing original draft (Equal), Writing - review & editing (Supporting). Nancy Scherer: Writing - review & editing (Supporting). Claire Schrock: Investigation (Equal), Writing - review & editing (Supporting). Ryan Seltzer: Methodology (Supporting), Writing - review & editing (Supporting). Carol Stoel-Gammon: Conceptualization (Equal), Writing – review & editing (Supporting). Lauren Thompson: Methodology (Equal), Writing – review & editing (Equal). Beate Peter: Conceptualization (Lead), Project administration (Lead), Writing – review & editing (Equal).

Data Availability Statement

The data sets generated during and/or analyzed during this study are available in anonymized form from the corresponding author on reasonable request.

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