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The feasibility of an online language programme delivered through music and the impact of dosage on vocabulary outcomes in young children with Down Syndrome.

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Abstract

Background: Few studies have explored the feasibility of online language interventions for young children with Down syndrome (DS). Additionally, none have manipulated dose frequency or reported on the use of music as a medium through which language and sign can be learned.

Aims: To 1) examine the feasibility and acceptability of an online language through music intervention for young children (1 – 3;06 years) with DS, and 2) compare effectiveness at two intervention dose frequencies.

Method: The study was carried out in two phases using a mixed methods design. *Phase 1:* qualitative data were gathered from parents to examine feasibility when implementing a video-based language intervention. *Phase 2:* Seventy-six families participated in an online language intervention at home. Effectiveness was examined comparing two groups, randomly assigned to a high and low dose frequency. The Down syndrome Education checklists (DSE) (combined) were the primary outcome measure. Process data were gathered to determine intervention acceptability in practice and to identify factors that would improve successful future implementation. Acceptability data were analysed with reference to the theoretical framework of acceptability (Version 2).

Results: Forty-three parents completed the phase 1 scoping questionnaire, five of whom took part in focus groups. Once weekly morning sessions were indicated as the preferred scheduling choice. Phase 2 quantitative data were analysed using Beta regression adjusted for baseline scores and indicated no additional benefit to receiving the higher dose. However, exploratory interaction models suggested that the efficacy of the high-dose intervention was higher (than low dose intervention) in participants with higher baseline DSE performance. Parents perceived the intervention to be effective and positive for the family.

Conclusions: The results add to our knowledge of real-world effective online-interventions and suggest a critical minimum language level is required for children with DS to benefit optimally from a higher intervention dose frequency.

Introduction

Down syndrome is the most common genetic cause of intellectual disability and is due to an extra copy of chromosome 21 (Abbeduto et al., 2007). It can also be the result of mosaicism, when only some cells include an extra copy of this chromosome, or translocation, when part of chromosome 21 attaches to another chromosome. Figures from Down Syndrome Ireland (2019 - 2022) estimate that about 137 babies are born with Down syndrome in Ireland each year, and international figures show that Ireland has one of the highest incidences of Down syndrome in the world (Loane, 2013). Children with Down syndrome experience a range of medical, physical, and developmental difficulties, and have relative strengths in the areas of visual memory and social understanding (Abbeduto et al.,2007; Burgoyne & Cain, 2020). The development of speech and language skills is one of the greatest areas of need, with receptive language typically better than expressive (Chapman et al., 2002). Moreover, research has identified that compared to other groups of people with similar levels of intellectual ability, those with Down syndrome show disproportionate language difficulties (Abbeduto et al., 2007; Frizelle et al., 2018; Yoder et al., 2014) which affects all aspects of their lives including their academic and social outcomes. The current paper therefore outlines the development of a language intervention for this group that capitalised on their strengths in visual attention and social engagement; incoroporated the facilitative role of music; and examined the effect of intervention dosage on the outcomes.

The language and communication difficulties of children with Down syndrome are apparent early in development (D'Souza, et al., 2017; Mason-Apps, et al., 2018). Research has

revealed early differences from typically developing children that have significant implications for language development. For example, children with Down syndrome have been found to have lower rates of engagement in object play than other children, including those with autism, and instead tend to be fixated on attending to their social partner (D'Souza, et al., 2017; Wright, et al., 2013). Although joint attention is in line with their developmental level (Hahn et al., 2018), children with Down syndrome have been found to have difficulty responding to joint attention with adults during play which limits their exploration of the environment aswell as their opportunities to learn from adults naming and talking about objects of interest (Abbeduto et al., 2007; D'Souza, et al., 2017; Mason-Apps, et al., 2018). Furthermore, although preverbal communication such as smiling and pointing follow the same developmental path as typically developing children (Martin et al, 2009), children with Down syndrome can be relatively passive in their communication compared to typically developing children of the same age (Mundy, et al., 1995). This can affect how parents interact with their children and result in parents using simpler vocabulary, fewer words, less grammatically complex utterances and a lower proportion of imitations in language input when compared to parents of typically developing peers (Zampini, et al. 2011). All of these factors result in a delay in the emergence of first words (circa 18-36 months old) in children with Down syndrome, compared to typically developing children, where first words usually emerge between 10 and 14 months (Abbeduto et al., 2011). This early delay has a cascading effect on the later development of all communication and language milestones including morphology, simple and complex syntax, and speech intelligibility (Eadie et al 2002; Chapman, 1997; Frizelle et al. 2018; Laws et al 2014; Naess et al., 2011; Martin et al 2009; Smith et al 2020). Although some language domains including receptive vocabulary and pragmatics, have been found to be less affected in this group (Laws et al 2015; Martin et al 2009), children with Down syndrome do exhibit weaknesses compared to typically developing children of a similar age (Smith et al, 2020). It

is also important to note that there is significant variability in the profiles of children with Down syndrome (Karmiloff-Smith et al., 2016) with some children showing considerable delay in their vocabulary development, relative to children with intellectual disability (ID) of non-Down syndrome (NDS) aetiology, matched for mental age (Warren et al., 2008).

The need for vocabulary interventions

Intervention is required to address the language difficulties exhibited by children with Down syndrome, and importantly, children with Down syndrome have been shown to respond to language intervention programmes which have resulted in long term benefits for language and communication (Smith et al., 2020; Seager et al 2022). For example, structured and guided opportunities for parents to interact with their young children with Down syndrome in naturalistic settings have been found to be beneficial for both parental interaction and child language (O'Toole et al., 2018; Seager et al., 2022). In addition, interventions that are co-delivered by parents and clinicians/researchers have the potential to result in positive outcomes for young children with Down syndrome (Seager et al,. 2022). However, evidence for the effectiveness of vocabulary interventions is scant, with few studies focussing directly on this aspect of language. Given that vocabulary affects the development of other language domains, and the reported gap between receptive and expressive vocabulary in children with Down syndrome, there is a distinct need to design and evaluate more vocabulary-focussed, robust interventions for this group (Smith et al, 2020). Seager and colleagues (2022) carried out a recent systematic review of speech language and communication interventions for children with Down syndrome (0- 6 years). Although they found over 1300 articles, only 11 met their inclusion criteria, five of which included vocabulary outcomes to measure intervention effects. The reported outcomes for vocabulary were mixed. For example, Girolametto et al., (1998) found no difference in vocabulary size between the intervention and control group, while Yoder et al (2014) found that children who received a higher dose intervention had a larger growth in

expressive vocabulary compared to those who received a lower dose. Moreover, although almost half the studies included vocabulary outcomes (reflecting an expectation that vocabulary will increase because of responsive teaching), only one study appeared to focus on the development of vocabulary directly. This finding was reinforced by Smith et al., (2020) who also note that few intervention studies for children with Down syndrome have focused specifically on this domain of language. Moreover, given difficulties with sustained attention, it is important that language interventions are engaging and that they build on relative areas of strength for people with Down syndrome, including their visuospatial processing, nonverbal communication, and socialisation skills (Buckley and Le Prevost, 2002; Abbeduto et al., 2007; Wright, et al., 2013). One way to build on these strengths and to engage young children with Down syndrome is to integrate music into language intervention programmes.

Benefits of incorporating music. Music has been extensively used as a treatment for a variety of developmental domains, including language and communication in children with disabilities (Cambell-Philips, 2020; Rushton & Kossyyaki, 2020; van Tellingen et al., 2023). Those with Down syndrome are reported to be particularly engaged with and communicative through music interventions . (Pienaar, 2012). Moreover, some have argued that as music and language develop from the same underlying cognitive processes involving auditory perception, attention, and memory (Zatorre, 2005), music can have positive impact on vocabulary and social development; learning the rhythmic patterns of speech; and communication. Moreno-Garcia et al., (2020) completed a systematic review of 19 papers that looked at the role of music on different developmental domains of children with Down syndrome. Five of the selected papers were relevant to the current study as they focused on communication development. One of these papers was a study by Vitoria-Gallastegi (2005), in which a musical education programme was reported to be associated with improved language comprehension and expression as well as related cognitive domains such as verbal memory, in a group of teenagers

with disabilities. Moreover, the impact was reported to be greatest for those with Down syndrome. A further report by Guy and Neve (2005) also documented the contribution of music therapy to the development of speech and language skills. A qualitative study by Pienaar and colleagues also included in the review, reported on caregivers' views of music therapy, working in a special school. Caregivers reported that children with Down syndrome were responsive to music and staff believed that it helped children to enhance their communication and develop social skills (Pienaar, 2012). Overall, Moreno-Garcia et al., (2020) concluded that the review found positive effects on the areas of social-emotional, motor, cognitive and communication skills but that most studies were of low quality; included a small number of participants; failed to describe the nature of the interventions in detail; used subjective outcome measures; and lacked an element of control in the study designs. Therefore, it would seem timely to develop more controlled intervention studies that incorporate the use of music.

Benefits of incorporating gestures/key word signing. Another important aspect of interventions tailored for children with Down syndrome is the use of non-verbal communication such as gestures. Gestures are a natural part of communication and are frequently observed in young children as they are learning to communicate verbally. However, many studies have noted how children with Down syndrome use gestures to communicate more frequently and for longer periods than typically developing children (Caselli et al., 1998; Franco & Wishart, 1995 & Steffanini et al., 2007). Children with Down syndrome have also been found to show a preference for gesture over vocal production during the early phase of lexical development, indicating a specific difficulty in expressive language (te Kaat- van den Os et al., 2015). Using gestures is one way to harness the visuospatial strengths of children with Down syndrome while simultaneously facilitating communication and spoken language.

One structured approach to the use of gesture is key word signing (KWS). KWS involves using manual signs to highlight the content/key words in a sentence, while

simultaneously using spoken language (Frizelle, 2019). Key word signs are intended to be iconic; focus only on the key information carrying words in the sentence; and do not mark grammatical forms such as possessive or past tense (Rombouts et al., 2020). KWS also takes advantage of the visual processing and imitative skills of children with Down syndrome (Vanvuchelen et al., 2011), increases time to process information and facilitate comprehension (Emmorey, 2002); increases children's attention (Frizelle et al., 2023); reduces working memory demands (Vanvuchelen et al., 2011); reduces language complexity in the input; and increases saliency of word boundaries (Rombouts et al., 2017). Indeed, interventions that include key word signing have been reported to be more effective than those in which spoken words alone are used (Wright et al., 2013; Neil & Jones, 2018). A recent study by Frizelle and colleagues (2023) investigated the effects of embedding signs in a shared book reading activity with young children with Down syndrome and found that parental use of sign increased children's participation (attention and initiation) compared to when signs were not included. te Kaat-van den Os et al (2015) completed a systematic review of 12 studies that investigated the role of gestures/KWS in acquiring vocabulary and grammar in young children with Down syndrome and found a predictive relationship between gestures and spoken word production. Özçalişkan and colleagues (2016) refined these findings further, highlighting the predictive nature of signs rather than simple deictic gestures. One study included in the te Kaat-van den Os review, explored the effect of a naturalistic intervention incorporating KWS on four young children with Down syndrome (aged 23-29 months) (see Wright et al., 2013). At the start of the intervention the children were using a maximum of one sign and had a spoken vocabulary of between two and 11 words. The intervention took place twice a week for 20 sessions and on completion, all children demonstrated an increase in the number of signed and spoken words used, along with a small change in joint attention. However, considerable individual differences were noted in the number of spoken words acquired, linked to individual children's imitation

skills as well as parental use of sign at home. The findings highlight the importance of a child's communicative partner learning to use and recognise KWS to respond optimally and provide language learning opportunities in their daily interactions. Consequently, best practice guidelines for preschool children with Down syndrome highlight the importance of including the use of KWS in early interventions, thereby facilitating the development of spoken words. In addition, they call attention to the need for parents to be trained in, or at the very least, consistently exposed to KWS techniques (Buckley & Le Prévost 2002).

Intervention Dosage

While rarely systematically manipulated, a key feature of effective interventions is the intensity to which they are delivered, such as the dose frequency and the total intervention duration. Previous research has noted that the effect of dosage is different depending on the child's underlying etiology (Fey et al.,, 2013). However, young children with Down syndrome seem to benefit from interventions with increasing intervention dose frequency. For example, while controlling for intellectual disability, Yoder et al. (2014) found that children with Down syndrome (mean age 22 months) had higher spoken vocabulary outcomes when they received a high dose frequency of the intervention (i.e. 5 x 1-hour sessions per week) compared to a lower dose frequency (1 x 1-hour session per week). Moreover, a systematic review and meta-analysis of communication interventions concluded that a large number of prompted opportunities for expressive language presented rapidly during frequent sessions work better for children with Down syndrome than a less intensive approach (Neil & Jones, 2018). Overall, the consensus in the literature is that high frequency interventions or those with a higher dose result in better outcomes for young children with Down syndrome (Smith et al., 2020; Seager et al., 2022). This is particularly significant in the context of a recent questionnaire completed by parents of adults and children with Down syndrome in Ireland (n= 557), indicating an average of only five public speech and language therapy sessions per year, as well as a mode

waiting time of 1 – 2 years to receive an assessment or treatment (Frizelle et al., 2021). However, limited Speech and Language Therapy (SLT)/ pathology (SLP) services are not unique to Ireland. A national questionnaire of children with Down syndrome in Norway also revealed that one third of children did not receive systematic intervention for their language difficulties (Naess et al 2017, as cited by Smith et al 2020) while just over 50% of the Irish sample received no intervention (Frizelle et al., 2021).

The current study

During the Covid 19 pandemic, Down syndrome Ireland (DSI) delivered a series of 10 40 minute online music sessions to a parent and toddler group. DSI is an organization which provides support and private therapeutic services for families of people with Down syndrome in Ireland. The music sessions were delivered live, once a week by the second author. The sessions focussed on the use of singing and musical instruments to engage preschool children with Down syndrome, and incorporated some use of KWS. When the planned series was complete, parents expressed the need for more and noted that the series was a very positive experience for all participants. DSI were in a position to fund another series of sessions but were interested in integrating language in a more systematic way. Consequently, the first (a speech and language therapist) and second (an academic musician) authors worked together to develop an online language through music programme. The intervention focuses on the development of vocabulary; incorporates the use of singing, common objects, and key word signing; and encourages child and family participation. As such it aims to address the difficulties children with Down syndrome experience with attention, while simultaneously capitalising on their strengths. The following research questions are addressed.

Phase 1

- What do parents consider important when implementing an online language through music intervention with young children with Down syndrome?

Phase 2

- What is the effect of dosage in an online language through music intervention on the vocabulary outcomes of young children with Down syndrome, with respect to their
    - understanding/use of signs?
    - imitation of words?
    - spontaneous use of words?
- How acceptable is the intervention to parents who participate?

Given previous literature indicating the advantage of high dose versus low dose frequency, we anticipated that those in the high dose frequency group (5 times a week) would have better outcomes than those in the low dose frequency group (twice a week), particularly with respect to the understanding/use of signs. We also expected this advantage to be evident in word imitation. However, given the short duration of the intervention and the immediacy of the post-intervention outcomes, we did not anticipate that any intervention effect /dose frequency advantage would translate into an effect for the spontaneous use of words.

Given that the design of the programme was informed by phase 1 of this research, we hypothesized that the intervention would be acceptable to parents overall.

Overall Design

This was a mixed methods study in which outcome and process data were collected. The study was carried out in two phases. In phase 1 qualitative methods (scoping questionnaire and focus group interviews) were used to ascertain what parents consider important when implementing on online language through music intervention, based on their previous experiences of music sessions or any other online programmes. In phase 2 we examined the effectiveness of our programme with respect to different dose frequencies. The results of this

study will therefore add to our knowledge of real-world effective online interventions, specifically levels of dosage required to effect change in the receptive and expressive vocabulary of young children with Down syndrome. Qualitative process data were also gathered in phase 2, to examine the feasibility of the approach in practice and to identify factors that would improve the probability of successful implementation.

Phase 1

Method

In phase 1 qualitative data collection methods and analyses were used. An online scoping questionnaire was initially used to gather data from a representative sample of parents on their previous experiences of online music sessions. This was followed up by a focus group interview with parents. A focus group was chosen as it provides an opportunity to gather more detailed data about participants' perspectives on a predetermined set of issues (Stewart et al., 2007), while at the same time allowing participants to interact with the ideas and comments made by other members of the group (Krueger, 2015). Ethical approval was given for this phase of the work by the Social Research Ethics Committee, University College (removed for anonymity).

Recruitment and Participants

Questionnaire participants were recruited through the Down syndrome Ireland (DSI) members database. The last author emailed a link to the scoping questionnaire, to all parent members who had a child with Down syndrome between 1 and 4 years-old (approximately 390 parents). Forty-three questionnaire responses were recorded through Qualtrics At the end of the scoping questionnaire parents were given the option to be part of a focus group to discuss in further detail, how the language through music sessions might run. They were asked to email the last author at Down syndrome Ireland if they were interested in taking part. Five parents expressed an interest in taking part and were included in the focus group. The 5 parents

represented different parts of Ireland and all were mothers of children with Down syndrome aged between 9 months and 2;09 years. Four of the five had taken part in the previous online music sessions. Questionnaire participant demographic information is shown in Table 1.

Table 1. Questionnaire Participant Demographics

Primary Caregiver	**Yes**		**No**	
	n	**%**	***n***	**%**
	36	83.7	7	16.3
Relationship to the Child	***n***		**%**	
Mother	40		93	
Father	2		4.7	
Guardian	1		2.3	
Work Outside of the Home	**Yes**		**No**	
	n	**%**	**n**	**%**
	27	62.8	16	37.2
Part-Time/Full-Time	***n***		**%**	
Full-Time	14		50	
Part-Time	14		50	
Part-Time <20 hours	8		57.1	
Part-Time >20 hours	4		28.6	
Self Employed	1		7.1	
Career Break	1		7.1	

Procedure

Scoping Questionnaire. A scoping questionnaire (delivered online) was developed by the first, second, fifth and last author, all of whom have significant experience developing interventions for children with Down sydnrome. The team discussed the main feasibility points of interest for the planned intervention and developed the questions accordingly. Four questions were closed multiple-choice questions and the other six questions were open ended allowing for a free text response. Assuming that not all responders would have attended previous online/music sessions, questions were phrased in the conditional as well as the past tense. Parents were asked what did/ did not work well in previous sessions; what helped or would help them to remember to sing the songs with their child; what helped or would help them learn the signs in the songs; and what did or would help them to keep their child engaged in the

session. Multiple choice questions focused on preferred method of online access and dosage preferences (e.g., session length and frequency). The scoping questionnaire content and layout is shown in Figure 1 in supplemental material. It was distributed using the Qualtrics platform and responses were collated by the second author.

Focus group. The focus group was scheduled in the evening time in December 2021. It was hosted on line by the last author, using the audio and web conferencing platform, Zoom. The link for the group was established with password protection and a waiting room to verify individuals attempting to join. The focus group was moderated by the last author who admitted each participant from the waiting room at the start of the session. The session was video and audio recorded through zoom. A semi-structured format was adopted. A series of questions, informed by the scoping questionnaire responses, were developed by the team. Key areas of focus included - what would be optimal programme time frames (overall duration, session frequency, session length)?; what did and did not work well in previous music sessions and why?; what helped to learn the signs?; what helped to remember to sing the songs between sessions?;; and what helped to keep children engaged in previous sessions?. At the beginning of the session the first author gave a brief introduction explaining the aim of the focus group and setting the context for the planned intervention. The remainder of the group was structured around the series of predetermined questions. Participant responses were captured by the Zoom audio recording and were transcribed by the second author.

Data Analysis

Scoping questionnaire and focus group data were analysed by the third author, a graduate with experience analysing qualitative data, but who had no involvement in the previous online music sessions or with the planned intervention. Free text scoping questionnaire data and focus group data were analysed qualitatively using a conventional content analysis (Hsieh and Shannon, 2005), while dosage preferences were analysed

descriptively (e.g. through frequency counts). Transcription of the focus group audio was available through the zoom recording. Focus group transcripts and free text scoping questionnaire responses were transferred to NVivo and segmented by question item. Data were read repeatedly by the third author to achieve immersion and a sense of the whole data set. Data were then segmented into units reflecting "the smallest amount of information informative by itself" (Vaughn et al., 1996, p. 106). Following segmentation the data were coded under each of the scoping questionnaire question headings. To increase trustworthiness all data and coding were discussed with and reviewed by the first author.

Results / Discussion

Because the focus group discussion centred around similar questions to those in the scoping questionnaire, no new themes emerged. Therefore we present the free text and focus group data together in our results. Responses to questions regarding dosage are shown in Table 2.

Table 2. Timing and Dosage Preferences

Preferred Time of Day	***n***	**%**
Mornings	24	55.8
Afternoons	8	18.6
Evenings	11	25.6
Nights	0	0
Preferred Length of Session	***n***	**%**
1 Hour	1	2.3
40 Minutes	7	16.3
30 Minutes	29 6	67.4
15 Minutes		14
Preferred Length / Frequency Combination		
	n	**%**
1 Hour Once a Fortnight	5	11.6
40 Minutes Once a Week	15	34.8
20 Minutes Three Times a Week	10	23.3
10 Minutes Three Times a Week	2	4.7
A Different Combination Not Specified	11	25.6

As shown in Table 2 the majority of participants stated a preference for intervention sessions to take place in the morning (56%) and a significant majority chose 30 minutes as the preferred session length (67%). Note, we believe that parents interpreted this question in the context of music sessions that would incorporate a variety of activities, held online at a set time each week (rather than pre-recorded music videos). There was considerable variation in the preferred session frequency / length combination, with most choosing 40 minutes once a week (35%); about a quarter of parents (23%) choosing 20 minutes 3 times a week; and a similar number choosing the option 'different combinations not specified'. Differences in session frequency preferences (with respect to parent child interaction therapy) have been previously reported, with some studies reporting a frequency preference of once weekly (Carr, 2011) and others preferring fortnightly sessions (Shamsudin et al., 2021). When creating the scoping questionnaire we were unsure about whether our online intervention would entail live sessions each week or pre-recorded videos, as our decision would be informed by parent preferences. However, given the different dynamic in each of these contexts it would have been preferable to present parents with both options and ask for dosage preferences with respect to each specific context.

What worked well in previous music sessions and why?

Parents stated that keeping sessions short (although they did not specify an optimal time) as well as the use of props, toys, instruments and colourful backgrounds worked well in previous sessions. They also highlighted the importance of making the session interactive where "All little ones were encouraged to sing along and sign." Using well known songs and lively music" was also deemed important, as shown in the following quote- "The use of well know song tunes or nursery rhymes [but] with new words to complete tasks such as brushing teeth/ hair has been a game changer for us and our little one". The need for children with Down syndrome to be active particpants in order to increase attention and maximise potential intervention effects has

been previously highlighted in the empirical literature (Burgoyne & Cain, 2020; Frizelle et al., 2023).

What did not work well in previous music sessions?

Attending to a screen and screen size were highlighted as potential issues compared to engaging with an intervention in person - "It's hard to do it online as my babies attention span isn't great." Distractions in the home were also stated as problematic. Some parents indicated that previous sessions went on for too long, were "…trying to pack too much into one session" or lacked active engagement/interaction with children. The timing and length of sessions were also noted as problematic, with one parent noting that fortnightly sessions did not work for them, causing them to lose momentum between sessions. This is in contrast to previous literature where fortnightly sessions (with respect to parent child interaction therapy) were the preferred choice (e.g,. Shamsudin et al., 2021). This preference difference may simply be a reflection of individual differences but is also likely influenced by the convenience of online sessions, where parents are not required to travel or to make supervision/baby-sitting arrangements for siblings.

What helped to learn the signs?

Repetition and the ability to be able to play back recorded sessions were considered by many parents to be important for learning signs. The benefits of repeated exposures to words/signs for those with a language disorder (including those with Down syndrome) is well reported in the intervention literature (Storkel et al., 2017; Frizelle et al., 2023) and it is therefore not surprising that the ability to playback sessions and therefore increase the number of exposures to sign/word pairings, was deemed advantageous. Parents also valued being sent either written information or video clips of the signs in advance of the sessions. One parent noted that the introduction of a small number of individual signs during each session was helpful to their learning and made it easier to remember new signs. Finally, the use of objects of reference/

visual aids and active interaction with the facilitator were noted to be helpful for learning signs. Given the relative visual memory strengths of people with Down syndrome (Rowe, Lavender & Turk, 2006) the reported benefits of visual aids is also unsurprising.

What helped to remember to sing the songs between sessions?

Twenty-five percent of parents noted that when songs were linked to daily activities and could be incorporated into their family routine, it was easier to remember to sing them - "Practice as part of our daily routine e.g. at mealtimes.....build it into part of our routine". This was in keeping with that reported in the systematic review completed by O'Toole et al., (2021), in which 67% of studies highlighted the importance of integrating therapeutic activities into daily routines.

Twenty percent of parents suggested that the use of familiar or catchy songs were easier to remember to sing - "I remembered if they were already familiar songs. Or if they were new songs that were catchy and simplistic/relatively few signs to do." This is in keeping with Popova (2019), who emphasises that the catchiness of a song is dependent on its relationship to known familiar tunes. In addition, she highlights that if a new song is presented in a previously experienced musical context, the brain is more receptive to the song and will retain its contents, thereby encouraging the person to sing along.

As was the case when helping to learn the signs, the ability to play the songs back during the week or a written aid to refer to was considered helpful. Ten percent of parents stated that they found it difficult to remember to sing the songs between sessions because "(fortnightly) sessions were too far apart to remember".

What helped to keep children engaged in previous sessions?

Over half of the parents noted the importance of interaction, communication with the facilitator, and fun and lively sessions for their children (56%). A considerable proportion (44%) also commented that using props and other visual aids helped their children to engage with the

sessions - “Visual puppets, facial expressions, music and laughing”. Other parents (24%) stated the use of songs would help to get their children’s attention, particularly if they already enjoyed music. A small group of parents (12%) noted that session length was an important factor when trying to keep their child engaged, favouring shorter sessions and “short simple songs”.

In summary, the key points of parental feedback that were brought forward from stage 1 to inform the design of the language through music programme delivered in stage 2 were as follows –

- Delivering the intervention through video, to allow parents to play back the recorded sessions at a time that was convenient to them and their children
- Keeping video lengths relatively short – (intended to be no greater than 20 minutes)
- Using props (objects of reference) toys and colourful backgrounds
- Making the session interactive such that each video incorporated a section to encourage the participation of those watching
- Using well known and lively song melodies with song vocabulary adapted to link with daily activities.

Intervention Development and Content

The language through music programme is an online 12 week programme of music videos developed by the first, second and last authors. The first and last authors are speech language pathologists and the second author a musician, all with significant experience working with, and developing interventions for children with Down syndrome. The programme combines active participation and evidenced based language learning techniques, such as repeated exposures to target vocabulary, the use of objects of reference and key word signing, with songs that have simple melodies and are therefore easy to remember. The programme development was also informed by parent feedback from phase 1 of the project and past experience of the team when effectively engaging young children with Down

syndrome. From a musical perspective, a range of well and lesser known melodies have been included. Song keys have been chosen to encourage adults to sing the songs, accounting for the fact that adults tend to have a lower tessitura (general range of pitch) than children (Sarrazin, 2016)). With respect to language, song lyrics have been altered to reflect vocabulary that is functional for young children; can be easily aligned with daily routines; and reflect words that are included in the Down Syndrome education checklists. For example, the lyrics in the song 'Baby Shark', have been changed to *Brush your teeth doo doo doo doo doo doo, Take a bath doo doo…., Go to sleep doo doo etc.* Each song is sung accompanied by Lámh the Irish key word signing system. There are 12 videos in total i.e. one video per week. The first eight videos are centred around different themes including *people, food, animals and bedtime*. The final four videos are a composite of the previous eight videos.

Each video includes two songs, presented three times- once where the song is sung with signs, once with sign and appropriate objects that represent the vocabulary in the songs and once with signs and objects but where words are omitted to facilitate parents' and children's participation by inserting the missing word/sign. An example of one of the videos can be found here (https://youtu.be/I_i18KILFrg).Video lengths range from 11.35 - 20.41 minutes/seconds (M = 17 minutes, 29 seconds). Songs range from 1.33 – 3.12 minutes/seconds in length, (M = 2 minutes and 21 seconds). Although the number of exposures to target words is controlled somewhat by the structure of the song, the presentation of vocabulary (and the accompanying signs) through song, results in a controlled dose of word exposures in each video. For example in the second video (animal theme) 15 words are targeted in song one, two items *go* and *zoo* are presented 42 times and all other words are presented 6 times. While in song two, 12 words are targeted, two items *washing* and *clothes* are presented 30 times, five words *here, there, down* and *goes* 15 times, and each animal, *elephant, lion, monkey bird, frog* is presented three times. Other videos such as video 3 (bedtime theme) allow for a more even dose of exposures,

where with the exception of one word, *sleep*, all other vocabulary items across both songs are presented 12 times. The minimum number of targeted words in each song was 5, and a mean of 12. The vocabulary list and the corresponding dose for each song and video are given in Table 1 in the supplemental material.

Phase 2

Method

In phase 2 effectiveness of the intervention was examined comparing two groups, of participants who were randomly assigned to a high (5 times a week) and low (twice a week) intervention dosage. Programme acceptability was examined through the development of a post-intervention questionnaire. Ethical approval for the study was granted through the Clinical Research Ethics Committee of the X (removed for anonymity) Teaching Hospitals.

Participants

Participants were recruited through the Down syndrome Ireland (DSI) members database, the DSI newsletter and social media. The last author emailed all parent members who had a child with Down syndrome who would be between 12 and 42 months old at the time the intervention would run. Our initial inclusion criteria were that children had a diagnosis of Down syndrome; were exposed to English in the home; and were between 12 and 42 months old. Seventy-six parents who expressed an interest in participating met our *prima facie* inclusion criteria. An additional 3 parents whose children were not yet 12 months old at the start of the intervention (but who were 12 months during the intervention), expressed an interest in taking part and were included in the intervention and the evaluation.

Outcome measures

Child baseline and outcomes. The Down Syndrome Education (DSE) checklists 1 and 2 (Down syndrome Education International, 2012) were the primary outcome measure. The checklists (based on the McArthur Bates Communicative Development Inventories; Fenson, et

al., 2007) are a parent report measure and account for the first 127 (checklist 1) and 340 words (checklist 2) understood and used by typically developing children (Down syndrome Education International, 2012). The DSE checklists document both receptive and expressive language and include five categories, Understands; Understands and signs; Says words in imitation; Uses words spontantously; and Understood by an unfamiliar listener. The checklists differ from the McArthur Bates Communicative Development Inventories in that they include both key word signing and words spoken as indicators of expressive vocabulary.

Acceptability Outcomes. A post-intervention questionnaire was developed to ascertain if the programme was acceptable to parents. The questionnaire questions were based on the theoretical framework of acceptability version 2 (TFA v2) (Sekhon et al., 2017). This framework includes seven component constructs designed to reflect the extent to which those delivering and or receiving a healthcare intervention consider it to be appropriate. The seven constructs are Affective Attitude; Burden; Ethicality; Intervention Coherence; Opportunity Costs; Perceived Effectiveness; and Self-efficacy. Additional questions outside of this framework asked parents to identify their three favourite songs and why; what would they change about the programme; and to rate on a sliding scale specific programme features in terms of their level of importance (i.e., weekly check-in sessions, frequent access to new songs/ signs, linking in with other parents online). The questionnaire was piloted with four parents and no changes were recommended. The post-intervention questionnaire questions are shown in Table 2 in supplemental material.

Procedure

Recruitment. A brief outline of the study along with details of two planned information evenings were posted in the DSI online newsletter. An email was also sent by the last author to all DSI members who had a child with Down syndrome between 12 and 42 months old, informing them of the study. The email included a consent form and a comprehensive

information sheet with contact details for the last author. Parents were invited to contact the last author if they planned to attend one of the information evenings; if they had any further questions; or if they were interested in participating. The two recruitment information sessions took place in August and September 2022. The first, second and last authors were present at each session. Information sessions were held online in the evening using the web conferencing platform, Zoom and scheduled on two different weeks to facilitate parents' attendance. Ten parents attended the August session and 36 parents attended the September session. The sessions were 30 minutes long and began with the first author giving a brief PowerPoint presentation outlining what would be involved in the study. The remainder of the session gave parents the opportunity to seek clarification on any aspect of the intervention or the evaluation that was unclear. Inclusionary criteria for the study were discussed and it was made clear throughout the session that parents could sign up for the intervention without being involved in the evaluation.

Allocation. Following consent to participate a computerized random number generator was used to assign participants to one of two treatment groups – a high dose group in which parents were asked to watch the weekly video 5 times a week and a low dose group where they were asked to watch the weekly video twice a week. Following randomisation each parent was emailed to inform them which group they were assigned to.

Intervention. A unique video link was emailed to parents on the Friday of each week for 12 weeks, from mid-September to December 2022. Depending on intervention arm parents were asked to watch the weekly video either five times (high dose) or twice (low dose) per week. Because treatment allocation was random, we were aware that there was a potential ethical issue regarding intervention dosage. For example, some parents who were assigned to the low dose group may have wanted to watch the videos more often than assigned, either because they believed it would be beneficial or because their child wanted to engage with the

video more often. Similarly, we were aware that some parents may have found it difficult to sustain five doses per week over the 12 week period. Consequently, we asked parents to log in a weekly diary the number of times that their children watched the videos each week and this would allow us to account for variation from treatment allocation. In order to encourage participation and peer support, parents were also invited to a Zoom meeting each Wednesday. Three time slots were allocated (morning, afternoon and evening) to facilitate parental attendance. In these groups parents discussed what was / was not going well for that week; how/whether their child was engaging with the programme; and perceived benefits of the programme. Attendance varied from week to week and all time slots were availed of.

Outcomes.

Child Outcomes. All pre- and post-intervention DSE checklists were completed by parents online. At the end of week 12, parents were emailed a link to the checklists which were administered using the Qualtrics platform. Baseline and post-intervention vocabulary measures were completed within 3 weeks of the programme commencement and completion.

Programme Acceptability. The programme acceptability questionnaire was also administered through Qualtrics. On the final day of the programme the first author emailed participants with a link to the questionnaire which remained open for 4 weeks.

Data Analysis

The three end-of-study outcomes were the proportions of combined DSE1 and DSE2 checklist words that the participant could respectively *Understand and Sign*; *Imitate*; or *Use Spontaneously*. Between-group (high-dose vs low-dose) effects were estimated using beta regression with adjustment for the outcome measured at baseline (Ferrari SLP, Cribari-Neto F. 2010; Simas AB, Barreto-Souza W, & Rocha AV, 2010). Beta regression models were also used to explore a baseline-by-group interaction, addressing the question of whether the impact of the intervention would be affected by the participants' language level, measured at baseline.

Finally, we similarly estimated the association between the actual dose of the intervention received, as reported by participants, and outcomes. Effect estimates from the beta regression models were reported as odds ratios with 95% confidence intervals and p-values. Regression diagnostics were checked using standard approaches. All quantitative analyses were conducted using the R Project for Statistical Computing (version 4.2.0; R Core Team, 2020). Beta regression models were fit using the betareg package (Cribari-Neto & Zeileis, 2010).

Qualitative data was analysed with respect to the Theoretical Framework of Acceptability (Sekhon, 2017). Post-intervention questionnaire responses were transferred to NVivo, segmented by the question items addressing each construct. Data were read repeatedly by the third author to achieve immersion and were then coded under each of the question headings. To increase trustworthiness all data and coding was discussed with and reviewed by the first author.

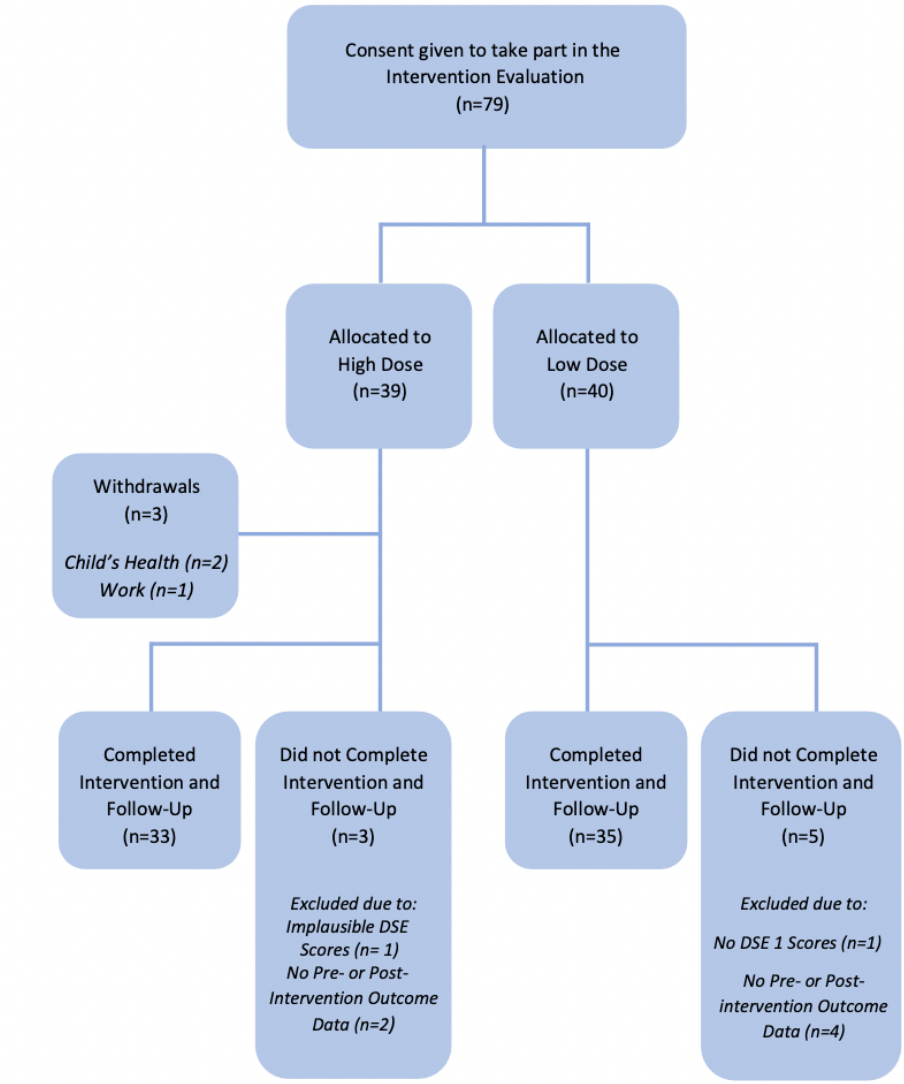
Results

Child outcomes

From the 79 children for whom consent was given to take part in the intervention evaluation, three were withdrawn prior to the intervention commencing. Two were withdrawn because of their health and one due to parent work commitments. In addition, there was no outcome data (pre or post) for six participants, one participant was missing post intervention scores for DSE checklist 1, and one participant was removed on the basis of invalid DSE scores (where the parent had indicated that their child understood and signed all words on both checklists).

This left a total of 68 participants included in the Phase 2 intervention evaluation, with 35 and 33 in the low and high dose groups respectively. The flow of children through the evaluation is shown in Figure 1.

Figure 1: PRISMA diagram of participants through the Intervention



Participants' characteristics at baseline are provided in Table 3. Child participants ranged from 9 to 45 months of age with similar proportions of males and females. Four participants came from bilingual families, and most had parents who had completed at least a 3rd level degree (63%). Participant characteristics were generally similar between the two groups, with the notable exception of baseline DSE scores, for which the low-dose group tended to have higher scores. This was particularly evident for the proportion of words that parents reported their children could *Use Spontaneously* (Table 3). The actual dosage

participants reported was generally similar to their assigned dosage with a median [IQR] of 2 [1.94, 2.13] mean doses-per-week in the low-dose group, and 4.7 [4, 5] in the high-dose group.

Table 3. Baseline Characteristics for Phase 2 participants

		Group		
	N	**Low dose, N = 35**[1]	**High dose, N = 33**[1]	**p-value**[2]
Age (months)	68	26 [17, 34]; (9 to 45)	21 [14, 28]; (9 to 44)	0.2
Sex	68			0.8
Male		16/35 (46%)	16/33 (48%)	
Female		19/35 (54%)	17/33 (52%)	
English language	68			0.3
bilingual		1/35 (2.9%)	3/33 (9.1%)	
yes		34/35 (97%)	30/33 (91%)	
Highest level of education	66			0.2
Primary school		0/33 (0%)	0/33 (0%)	
Lower secondary		0/33 (0%)	0/33 (0%)	
Upper secondary		3/33 (9.1%)	2/33 (6.1%)	
PLC course		0/33 (0%)	0/33 (0%)	
3rd level non-degree		2/33 (6.1%)	5/33 (15%)	
3rd level degree		14/33 (42%)	7/33 (21%)	
Postgraduate		14/33 (42%)	19/33 (58%)	
Adaptive behaviour	58	79 [73, 85]; (54 to 89); n = 30	79 [72, 83]; (42 to 99); n = 28	0.8
Communication	58	78 [67, 85]; (39 to 98); n = 30	78 [66, 84]; (24 to 92); n = 28	>0.9
Daily living skills	58	80 [69, 85]; (45 to 95); n = 30	79 [71, 84]; (39 to 106); n = 28	0.8
Socialisation	58	88 [80, 92]; (56 to 110); n = 30	86 [81, 95]; (49 to 101); n = 28	0.7
Understand and sign	68	0.02 [0.00, 0.10]; (0.00 to 0.52)	0.00 [0.00, 0.04]; (0.00 to 0.20)	0.14
Say word in imitation	68	0.00 [0.00, 0.02]; (0.00 to 0.37)	0.00 [0.00, 0.01]; (0.00 to 0.14)	0.085
Use word spontaneously	68	0.00 [0.00, 0.01]; (0.00 to 0.19)	0.00 [0.00, 0.00]; (0.00 to 0.14)	0.014

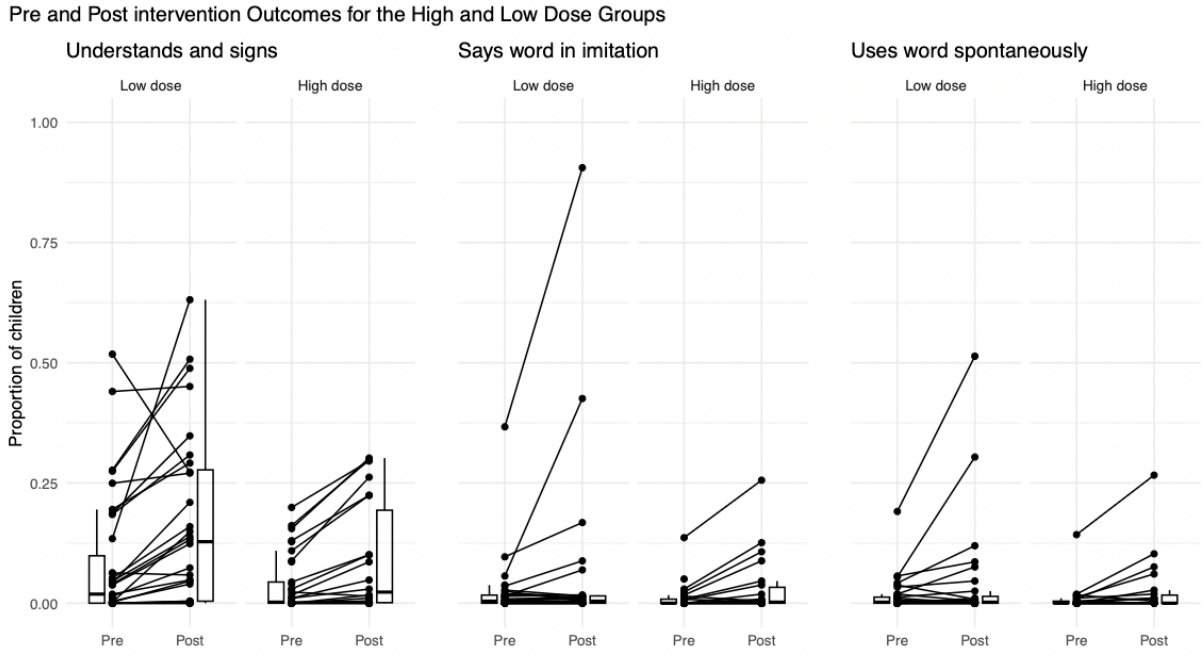
[1] Median [IQR]; (Range); n/N (%); n = N

[2] Wilcoxon rank sum test; Pearson's Chi-squared test; Fisher's exact test

In phase 2 of our research we aimed to ascertain the effect of dosage on the vocabulary outcomes of young children with Down syndrome, with respect to their 1) understanding/use

of signs 2) imitation of words and 3) spontaneous use of words. Of the 68 participants in our analysis sample, 50 had valid measurements for all three outcomes, the proportions of DSE checklist items that a participant reported being able to *Understand and Sign*, *Imitate*, and *Use Spontaneously*. Generally, participants in both groups experienced improvements in these outcomes over the 12 weeks of the study intervention (Figure 2).

Figure 2.



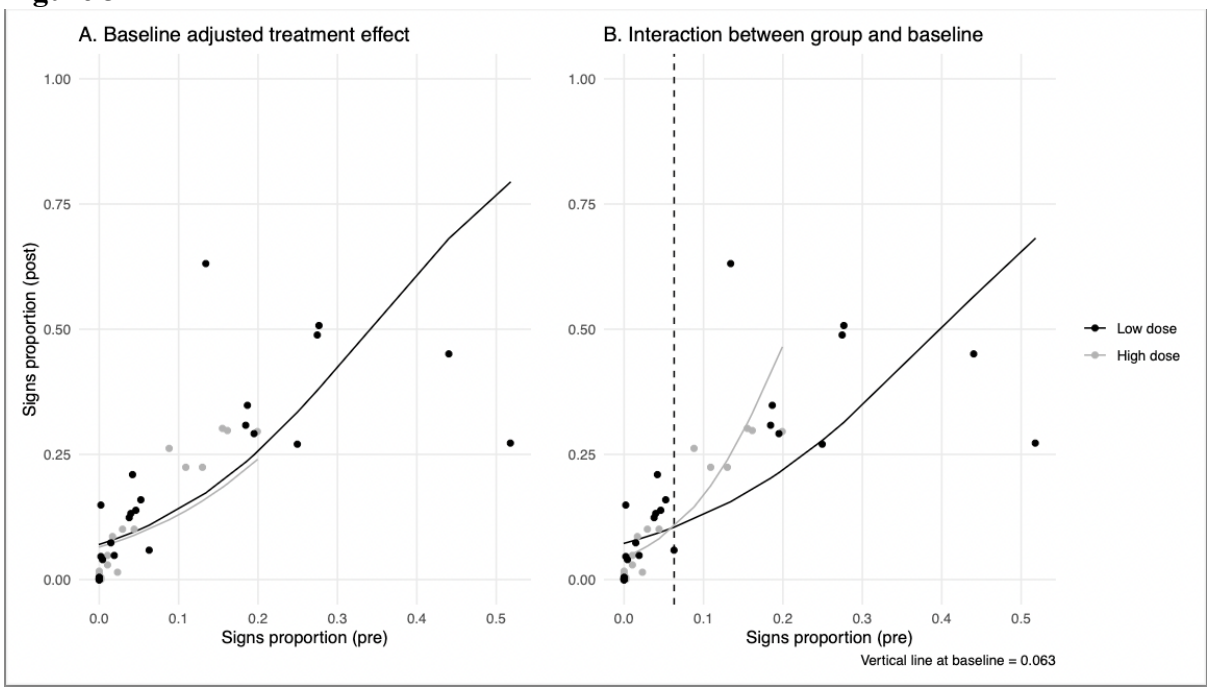
However, the estimated odds ratios (ORs) for the treatment effects comparing the high-dose to the low-dose group for each of the outcomes did not suggest any additional benefit of being allocated to the high-dose group (baseline-adjusted ORs of 0.92 (95% CI 0.54 to 1.57); 1.04 (0.63 to 1.71); and 1.02 (0.63 to 1.64) for *Understand and Sign* (Table 4, Figure 3A)*, Imitate,* and *Use Spontaneously*, respectively.

Table 4. Estimated odds ratios (ORs) for treatment effects comparing the high and low-dose groups for each of the outcomes.

	Understand and Sign			**Imitate**			**Use Spontaneously**		
Predictors	*OR*	*CI*	*p*	*OR*	*CI*	*p*	*OR*	*CI*	*p*

High dose (vs low-dose)	0.92	0.54 – 1.57	0.758	1.04	0.63 – 1.71	0.885	1.02	0.63 – 1.64	0.941
Baseline	1.08	1.06 – 1.10	**<0.001**	1.22	1.17 – 1.28	**<0.001**	1.26	1.21 – 1.31	**<0.001**
Observations	50			50			50		
R^2	0.441			0.504			0.513		
AIC	-159.203			-300.937			-327.478		

Figure 3



Additional adjustment for participant age did not appreciably alter these results. An exploratory interaction model including baseline DSE performance and treatment arm (high/ low dose frequency) was then completed. The model suggested that, for *Understands and Sign* (but not for the other two outcomes, where performance was very low relative to *understands and signs*), the efficacy of the high-dose intervention (relative to low dose) was higher in those participants with higher baseline DSE performance (Supplemental Table 3). More specifically, baseline-adjusted outcomes were higher in high-dose group than the low-dose group among

those with a baseline *Understands and Signs* proportion of at least 0.063 (Figure 3B), though there is some uncertainty in this number, with a bootstrapped 95% CI of 0.01 to 0.15. A final exploratory model including baseline DSE performance and reported weekly average dose suggested that an increase of one in a participant's weekly average dose, adjusted for baseline and group, was associated with improved outcomes, though these estimates had wide CIs (adjusted ORs of 1.20 (95% CI 0.96 to 1.50); 1.08 (0.86 to 1.34); and 1.09 (0.89 to 1.34) for *Understand and Sign, Imitate,* and *Use Spontaneously*, respectively).

Programme Acceptability outcomes

Fifty-eight of the 68 participants responded to the post-intervention questionnaire and were analysed. Overall, participants considered the intervention to be an acceptable online language through music intervention for young children with Down Syndrome. We have summarized our findings below using each of the constructs from the theoretical framework of acceptability (Sekhon, 2017).

Affective attitude. How an individual feels about the intervention: Fifty- eight parents completed this question. Parents commented on how much they and their children enjoyed taking part and benefited from the programme in their everyday lives. "I have really seen a difference with [child]. She engages better with interactive play and is definitely understanding more words since we have started signing more with her". They also noted benefits for the wider family stating "As a parent I learned a lot of signs that I actually remember through the song repetition….", "….great way for parents and siblings to learn as well as the kids. My 5- and 3-year-old loved them". One parent expressed a negative view of the programme stating "This programme did not suit my child. He needed to hear the songs more often than twice (a week) and to focus on 2 maybe 3 words." A small number of parents were also concerned that their child was too young to optimally benefit from the programme.

Burden. The perceived amount of effort that is required to participate in the intervention: From a total of 56 responses, 42 participants (75%) reported that the amount of time and energy needed to participate in the programme worked for them, while 14 participants (25%) reported that the time and energy burden was an issue. The time commitment associated with taking part in the programme was highlighted primarily by those who were assigned to the five times a week condition. Video length was also raised as an issue and a potential adaptation to a future iteration of the programme, with one parent stating "I think it would be too taxing for a young child to spend more than 15 mins more than twice a week watching these videos. If they were shorter, then doing it more often would not be an issue I think". In contrast, a smaller group of parents stated that they would have liked to watch the videos more frequently. "If this was not a test program with set limits on times to watch we would have watched it daily or more. My son kept asking for song in Lámh". Parents also referred to the impact of other commitments such as working outside the home, stating, "I am working full time at the moment and trying to get her to watch videos when she was tired in the evening was difficult". Finally, unforeseen life events such as illness, were reported to increase the burden for families trying to watch the videos a prescribed number of times each week.

Opportunity costs. The extent to which benefits, profits or values must be given up to engage in the intervention. From a total of 56 responses, 49 (87.5%) reported not having to give up anything in their daily lives/ routines to take part in the programme, while seven (12.5%) reported the opposite. Positive comments indicated the ease with which it could be incorporated into their daily routine, for example, "We generally were able to slot in the 15-20 mins to view the videos after our morning physio/OT routine. Delighted you have helped us add in some dedicated SLT time to our routine!". In contrast some parents noted that other forms of play were put aside to make time to take part in the programme. Most negative comments referred to finding time to attend the weekly support sessions rather than watching

the videos. Despite the three options to attend (morning, afternoon, evening) some parents found it difficult, due to trying to balance their own work schedules during the day as well as meeting the demands of their other children in the evenings.

Ethicality. The extent to which the intervention has a good fit with an individual's value system. Fifty-five (98%) participants agreed that the programme supported and encouraged positive aspects of their family value system, while one participant (1.79%) reported that this was not the case. Parents stated how taking part in the programme encouraged them to spend time together as a family while watching the videos and learning the signs and fitted well with families' interests - "We absolutely value communication in our family, and we LOVE music...a perfect fit".

The participant who stated that the programme did not fit with their family values reported that this was due to the amount of screen time the programme required. "Wouldn't normally allow 20 mins of screen time in one period for our child". This concern was also raised by one other parent who stated that while the programme did fit with their family values overall, "I try to minimise screen time for my kids, so it did feel strange putting a screen in front of my 20-month-old".

Intervention Coherence. The extent to which the participant understands the intervention and how it works. All participants stated that they understood the purpose of the programme and how it works, but only 14 of the 57 respondents elaborated further. Parents who did expand on the response noted the key components of the programme to be learning language and signs through repetition in music and song. One parent stated, "By singing the same rhymes every day and incorporating as many new signs a week as possible was a great way to learn signs through singing."

Perceived Effectiveness. The extent to which the intervention is perceived as likely to have achieved its purpose. Fifty-three of the fifty-seven respondents (92.98%) agreed that the

programme achieved its purpose, while four participants (7.02%) disagreed. Parents commented that the programme increased their child's spoken and receptive vocabulary as well as their signing ability -"She also has a greater understanding of some of the objects of content in the videos, like the animals etc which she previously wouldn't have had." In addition, they identified an improvement in their own competence using Lámh - "I am much more confident and consistent in my use of Lámh and this will have a long-lasting impact on my baby's learning over time". Another parent highlighted the effect of the programme on her child's sibling, stating "Our son loved the music, he isn't signing yet but our 2.5 yr old picked up signs & will help teach his younger brother". Those that did not think the programme had achieved its purpose noted engagement difficulties and believed that their child was either too young or that their child's communication skills were not developed sufficiently to benefit from the programme.

Self-efficacy. The participants confidence that they can perform the behaviour required to participate in the intervention: From a total of 57 participants, 54 (94.74%) stated that they feel confident to carry on with the programme themselves and noted that the availability of the videos would facilitate that. Participants stated that they intended to continue using the songs and learning the signs from the programme, either through repeated watching of the videos or singing and signing independently. They also commented on how the family friendly nature of the programme would help them to incorporate it into their daily routines. "[…] We often use Lámh signs when singing to learn new signs and words, it is an enjoyable activity that everyone at home can get involved in".

Top three favourite songs. Fifty-three parents responded to this question. Over half the sample (n = 28) chose either 'Wheels on the bus' or 'Baby Shark' as one of their favourite songs. "Lets go to the Zoo" and "Down in the Jungle" were also popular choices. Interestingly, every song from the programme was chosen as a favourite by at least one participant, showing

the diversity in people's musical preferences. Participants favoured songs that were 'catchy'; easy to remember; familiar to them; and that reflected parts of their daily routines.

The relative importance of each part of the programme. From 57 responses, having frequent access to new songs and signs was rated highest by participants, with a mean score of 86.77 on a scale from 0 to 100 (SD = 18.29; Max = 100, Min = 4). The weekly check-in sessions (50 responses) and linking with other parents (45 responses) were rated similarly by participants, with mean scores of 54.40 (SD = 31.62; Max = 100, Min = 1). and 54.89 (SD = 30.28; Max = 100, Min = 0) respectively. While some parents loved the weekly check-ins and found them helpful and encouraging, others found themselves comparing their children to others and consequently felt guilty that they weren't doing enough for their child.

What parents would change about the programme. Based on 54 responses, the length of the videos was the aspect of the programme most parents believed should be changed. Some videos were considered too long which caused difficulties with respect to 1) the child's level of engagement/attention and 2) families finding time to watch them during the day. One parent stated that "…. a video of approximately 7/8 minutes would be more effective" and another that "….after 15 mins she was disengaged and even after 10 mins the value was probably well reduced at that stage". Some participants believed that the content of the videos was too complex for their children (food/drink songs cited as an example) and that a reduction in the number of new words and signs per video with increased repetition, would be helpful. In contrast other parents suggested a reduction in the amount of repetition as a way of reducing each video length. Parents also suggested using bigger, more colourful pictures and props to help sustain their children's attention, while others suggested that the use of props was distracting for their child and took their attention away from the signs. A few parents suggested developing different programmes that would be tailored to preschool children of different ages.

Finally, some parents stated that they would prefer to access the videos in a different way, using YouTube for example.

Discussion

Based on our clinical experience and the feedback given in phase 1 of this study we developed an on-line language programme delivered through music, which focussed on vocabulary development in very young children with Down syndrome and incorporated the use of KWS, common objects of reference and active participation.

In our first research question we explored the effect of manipulating the dose frequency of the intervention and hypothesized that those receiving a higher dose would achieve better outcomes. Our results reflected a more complex message. Despite randomization at the outset, those in the low dose group had higher DSE checklist scores at baseline than the high dose group, on each of the three outcomes of interest (*Understands and Signs*, *Imitate*, and *Use Spontaneously*). We therefore adjusted for this in our statistical analyses. Despite our statistical adjustment, and contrary to our hypothesis, our results indicated that being allocated to the high-dose group did not hold any appreciable benefit above and beyond being allocated to the low-dose group. However, there are a number of potential explanations that could account for our findings. Firstly, given the sample size, one could posit that the study was not sufficiently powered to detect a modest but meaningful benefit of being allocated to the high-dose group. Secondly, the intervention may not have been long enough for the cumulative benefit of the higher dose to show any effect in such young children. Thirdly, children enrolled in the study may have required a higher level of baseline language knowledge to benefit from the higher dose of intervention. This second explanation is supported by our exploratory analysis showing that those with a baseline of 6% on the combined DSE checklists in *Understands and signs* (approximately 28 vocabulary items) tended to benefit more from the high dose intervention.

This latter explanation accords with previous literature reporting inconsistent findings with respect to dose frequency manipulations, highlighting that more treatment is not always better for all children (Frizelle et al., 2021; Schmitt et al., 2016; Storkel et al., 2017; 2019; Yoder, Fey, & Warren, 2012). This inconsistency implies that the effect of dose frequency differs according to specific child variables (Fey et al., 2013). Our findings suggest that pre-intervention language levels is one such variable and is in line with previous work carried out by Yoder and Warren (2002) who found that the spoken language skills of children with intellectual disability (but without Down syndrome), facilitated a more favourable response to their intervention, than for the children with Down syndrome (who had lower language skills).

Our results are also in keeping with Vygotsky's theory of the 'zone of proximal development' (1978), in that children who had not yet learned to use words or to sign (or were signing minimally), required more scaffolding than was provided in the videos, to learn the signs and begin to use new words. In contrast, those who had a more substantial repertoire of words/signs pre-intervention, appeared to be capable of learning in a more independent manner and consequently the format of the intervention was sufficient to support them.

Other non-linguistic characteristics of children in the high dose group may also have limited their response to treatment. Lower pre-intervention language levels may also be indicative of poorer attention skills (Clibbens, 2001) which may become more problematic when the intervention is delivered online. Some studies have suggested a tendency for children with Down syndrome to avoid tasks that are difficult for them (Wishart, 1993). Children who did not yet have an understanding of a critical minimum number of words, may have deemed the task too difficult, disengaged more readily from the videos, and therefore were less likely to benefit from any increase in dose frequency. This disengagement from the videos was supported in our qualitative findings with reference to the very young participants (who are likely developmentally to have lower language levels).

Finally, although our participants were randomly assigned to two different dose frequency groups, there were practical issues for parents adhering strictly to these dosages and we anticipated that there would be considerable dose frequency variability within groups. While parent logs showed that the actual dosage reported was similar to the dosage they were assigned for most participants, there was some variability, with some parents in the low-dose group reporting more than 2 doses per week (mean 2.2; range 1 to 4.5), and some in the high-dose group reporting fewer than 5 doses (mean 4.7; range 1 to 6.5). It is likely that this variability in participant dosage affected our findings. However, we aimed to explore the optimal dosage for this intervention and therefore used an exploratory dose response model (using actual weekly average dose and DSE outcomes) to explore potential effects. We found that based on actual dose, higher dose (across all children) was associated with better end-of -study outcomes, though these estimates were non-significant and accompanied by wide CIs. Although we interpret this finding with caution (as there may also be other confounding factors at play), this was in keeping with our hypothesis and is in line with previous research by Yoder and colleagues (2014) as well as findings by Neil and Jones, (2018). Increasing the frequency of sessions results in a more intensive treatment programme and increased exposure to word/sign pairings. The posited mechanism of action is that increased consistent repetition, can induce an acceleration in learning rate over time, (Warren, Fey, & Yoder, 2007), at least for those who are at a level to derive the benefit of the repetition increase.

In our second research question we asked if the programme was acceptable to parents who participated and what they would change about the intervention to increase acceptability. Overall, parents enjoyed the programme and believed that it was beneficial for their child with Down syndrome, their siblings, and the parents themselves. Examples of child benefits include noting an increased understanding of words and signs, increased spoken vocabulary and an increased ability to engage in play and interaction. The value of including siblings in

interventions has also been highlighted in previous studies (for example Amsbary, 2019; Amsbary et al., 2020; Daczewitz, 2015; Pennington & Noble, 2009) as it helps parents to engage with and implement interventions. Given the limited public SLT/ SLP services for very young children with Down syndrome in Ireland (Frizelle et al., 2021) we anticipated a positive response to the programme overall, reflecting a feeling perhaps that 'some intervention is better than nothing'. However, parents provided both positive and negative feedback. For example, some parents assigned to the low dosage group believed that more than twice a week would be needed to effect change and other parents considered their children to be too young for the programme.

While most parents did not find the programme burdensome, some parents assigned to the high dose condition found it difficult to find the time to watch the videos five times a week and highlighted the difficulty of competing demands on their time, such as working outside the home. Time demands were also highlighted in a review of parent experiences of parent child interaction therapy by O'Toole and colleagues (2021), in which 16 out of 27 studies referred to the difficulties working parents had finding the time to commit to interventions. While the demands of an online programme, such as that presented here, could be deemed much less burdensome than parent children interaction therapy (which is usually delivered in person), it was interesting that some working parents did report a dose frequency of five times a week as challenging.

Most parents reported not having to give up anything in their daily lives/ routines to take part in the programme and some commented that it helped them to add dedicated SLT/ SLP time to their routine. Almost all agreed that the programme supported and encouraged positive aspects of their family value system. The importance of attending to family belief systems when developing an intervention has been raised in previous studies (e.g., Kinard, 2015; Mendez, 2019; Pennington & Noble, 2009). Only two parents in the programme raised

the issue of screen time as a concern. This was surprising given the WHO guidelines of zero screen time, (except for video chatting with family or friends) for children under two years. Perhaps this was because screen time in the context of the WHO guidelines is usually defined as something that is passive and not educational, rather than an intervention focused on vocabulary development.

All parents stated that they understood the programme and how it worked. This was not surprising as two online sessions, explaining the content and purpose of the programme, were given pre-intervention. Overall, parents thought the programme achieved its purpose, noting improved receptive and expressive vocabulary and signing ability in their children. However, there were a few parents who believed their children were too young to benefit. These children were outside our intended target age, but their parents were interested in participating in the programme, and we did not wish to exclude them at the outset. Following the completion of the programme, parents' belief that some children were too young is in keeping with our finding that children needed a critical minimum level of language at baseline to benefit optimally from the intervention. Almost all parents stated that they felt confident to carry on with the programme themselves and noted that the availability of the videos would facilitate that. They also noted that because many of the songs were based on daily routines, it would be easier to continue with the programme. As noted in phase 1 of this work, this was in keeping with that reported in O'Toole et al., (2021), in which 18 of the 27 studies reviewed, highlighted the importance of integrating therapeutic activities into naturalistic daily routines for families.

While some parents underscored the value of the weekly check-ins with participating parents, others found themselves comparing their child to other children, and felt guilty or frustrated that they were not doing enough. Varied reports on the value of parental peer support have been noted in previous studies, with problems more likely to emerge when children receiving the intervention are of mixed ability (see Fäldt et al., 2020; Senent-Capuz et al.,

2022). Given the heterogeneity of children with Down syndrome, creating an environment of support without competition is an ongoing challenge.

Finally, with respect to what parents would change about the programme, the length of the videos was most frequently raised as an issue, with some parents suggesting that a video length of approximately 10 minutes would be optimal. Ostensibly, this seems very much in contrast to the parental feedback we received at the outset, where 30-minute sessions were the most popular choice. However, as previously stated, we believe that parents interpreted this question in the context of attending a live session (involving several different activities) at a set time each week, rather than preferring 30-minute videos of songs. In addition, parents suggested a reduction in the number of new words and signs introduced per video, with increased repetition. The total number of unique signs introduced over the 12 weeks was 148 and ranged from 11 – 27 in a single session. This is in stark contrast to Wright et al., (2013) who introduced a total of 32 signs over the course of their 10 week naturalistic sign intervention. While the song repetitions facilitated a high dose of word/sign exposures per video overall (which has been shown to increase children's word learning), parent's believed that the increase in number of new signs each week was too rapid for some children and preferred a more gradual approach to the introduction of new sign vocabulary. In addition, there was significant variation in the number of exposures to each word/sign pairing per video (M = 12.3, Med = 9) with some words having as high a dose as 72. In future interventions we would reduce the number of new signs introduced over the full 12-week period and attempt to achieve a greater within-session dose consistency for each word /sign pairing, although word/sign dose is somewhat dictated by the type of song.

Limitations

This study was primarily designed as a feasibility rather than an effectiveness study and therefore we did not include a control group. Although participants in both high and low dosage

groups experienced improvments in outcomes over the course of the intervention, a control group would have allowed us to definitively ascertain whether both dose frequencies facilitated an increase in understanding/use of signs, compared to no intervention. In addition, although parent logs showed that the actual dosage was similar to the assigned dosage there was variability within groups, which may have masked somewhat the benefits of a higher dose.

Secondly, the children included in this study were very young and consequently we did not have other measures such as hearing, vision or phonological short-term memory. The age of the children also meant that any children with a dual diagnosis (such as Autism or attention-deficit–hyperactivity disorder, both of which are common in Down syndrome (see Oxelgren et al., 2017)), could not be identified. Any of these factors could have moderated children's response to the different dose frequencies.

Thirdly, our outcome measure was a parent report, and although it is considered one of the most valid and reliable measurement options for use with young children (Marchman et al., 2023), parents cannot be kept blind to the purpose of the intervention. This could have led to biased reporting or parents anticipating a result, particularly for the high dose group. Initially it was our intention to keep parents blind to the two dose frequencies, however the weekly check-ins and conversations between parents made this impossible.

Conclusion

Overall, our language through music intervention was deemed both feasible and acceptable and was rated positively by most parents. However, some minor changes were recommended with respect to the number of signs that were introduced weekly and the length of the videos. In addition, a dose frequency of 5 times a week was considered burdensome for some parents. Although our initial analysis suggested no additional benefit to being assigned to the high dose compared to the low dose group, further analysis revealed an interaction between dose frequency and baseline DSE performance, such that those with higher

performance at baseline benefited more from a higher intervention dose frequency. The results of this study add to our knowledge of real-world effective online-interventions and suggest that 1) a critical minimum language level is required for children with Down syndrome to benefit optimally from a higher intervention dose frequency and 2) an increase of one in a participant's weekly average dose was associated with improved outcomes. Clinicians should take this into consideration when designing vocabulary based/ signing interventions for children with different language abilities. In addition, this information can inform discussions with parents on intervention dosage, in the context of video based interventions at home. Further exploration is needed to ascertain the overall effectiveness of this intervention and the optimal dosage at different language levels.

Data availability: Data for this study are available at the following link https://osf.io/3c2sd/

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Supplemental Files

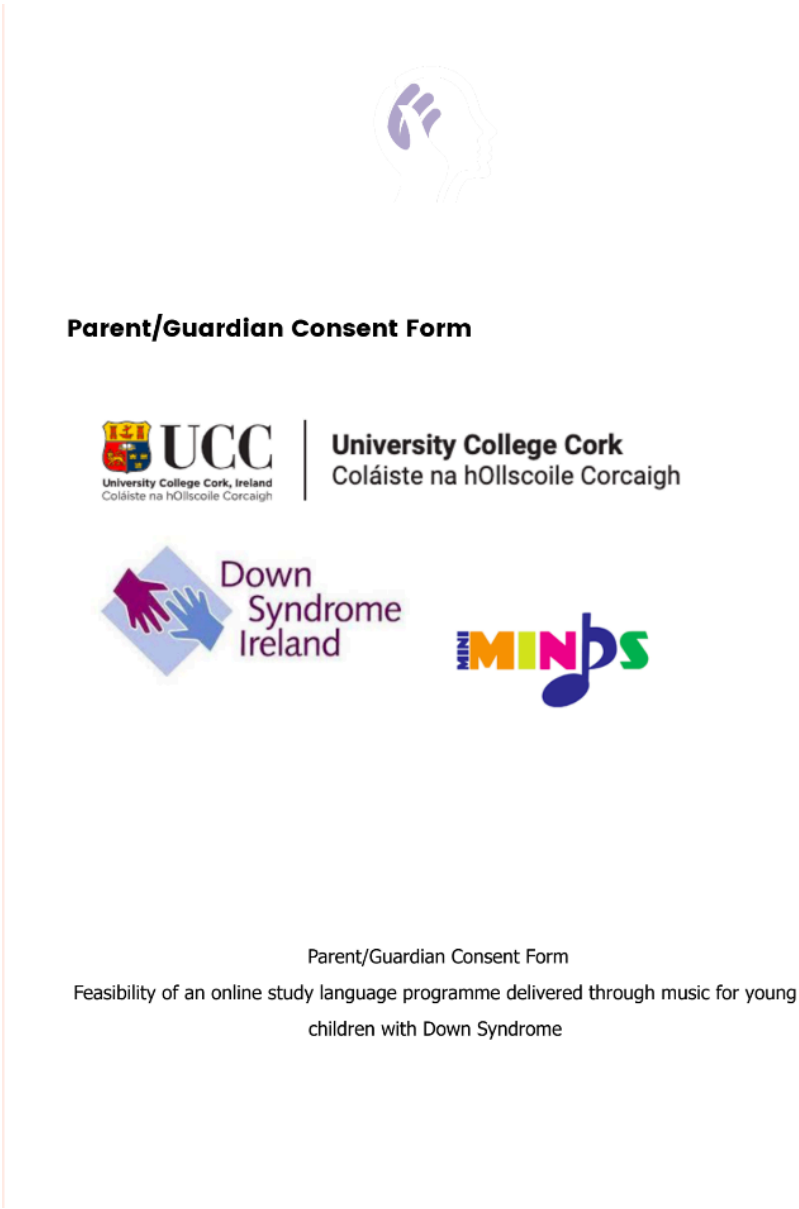
Figure 1 ***Supplemental*** **–** Scoping Questionnaire

Table 1 ***Supplemental*** **–** Vocabulary List and Corresponding Dose for Each Song

Table 2 ***Supplemental*** - Post-Intervention Questionnaire

Table 3 ***Supplemental*** -Exploratory interaction model of dosage with baseline DSE performance

Figure 1 ***Supplemental*** **–** Scoping Questionnaire



Parent/Guardian Consent Form

UCC
University College Cork, Ireland
Coláiste na hOllscoile Corcaigh

University College Cork
Coláiste na hOllscoile Corcaigh

Down Syndrome Ireland

MINDS

Parent/Guardian Consent Form

Feasibility of an online study language programme delivered through music for young children with Down Syndrome

The survey is anonymous and should take between 5 and 10 minutes to complete.







Parent/Guardian Consent Form

Feasibility of an online study language programme delivered through music for young children with Down Syndrome

The survey is anonymous and should take between 5 and 10 minutes to complete.

I have read and understood the details of the above study, and have had the opportunity to ask questions and discuss the study with others. I have received satisfactory answers to my questions.

Yes

I understand that the project has received ethics clearance through the Social Research Ethics Committee University College Cork, and understand who will have access to the data, how it will be stored and what will happen to the data at the end of the study.

Yes

I understand that participation is voluntary and whether I decide to take part or not will not have a bearing on my involvement in the program or any other services available from Down Syndrome Ireland.

Yes

I understand how to raise concerns or make a complaint. I understand that I can withdraw from the study up until the end of the intervention at which point my data will be anonymised.

Yes

To find out more about the study, please read the attached information sheet. You can also e-mail us at e.mcmullan@ucc.ie or p.frizelle@ucc.ie

Confirmation that my child is between 0 and 3 years

Yes

I am happy to take part in this survey

Yes

Date of Completion

	Month	Day	Year
Please Select:			

Survey

University College Cork
Coláiste na hOllscoile Corcaigh

Feasibility of an online language programme delivered through music for young children with Down Syndrome

Language Through Music Survey

If you have attended any previous music sessions online, what did you think worked well?

If you have attended any previous music sessions online, what did you think did not work?

How did you access the sessions

- ☐ Desktop
- ☐ Laptop
- ☐ Tablet/Ipad
- ☐ Phone

If you were to attend music sessions online, would you be willing to attend

- ○ Once a fortnight
- ○ Once a week
- ○ Twice a week
- ○ Three times a week
- ○ Other

preference

What time of the day would you like the sessions to be held?

- ○ Morning
- ○ Afternoon
- ○ Evening
- ○ Night

Do you work outside of the home? (it is helpful to have an understanding of your circumstances when interpreting your responses)

- ○ Yes
- ○ No

If yes please indicate whether it is full or part time

- ○ Full time
- ○ Part time

If you work part time please indicate how many hours per week

Are you the primary caregiver?

- ○ Yes
- ○ No

Please specify your relationship to your child?

- ○ Mother
- ○ Father
- ○ Guardian

If you attended our previous music sessions what helped you to learn the signs of the songs you were introduced to? If you haven't attended previous sessions, what do you think would help?

If you attended our previous music sessions what helped you to remember to sing the songs during the week? If you haven't attended previous sessions, what do you think would help?

What helped or would help keep your child engaged in the session?

The survey is anonymous, if you would like to be part of the focus group to discuss how the language through music sessions might run (how often, for how long etc.) please email Nicola Hart at Down Syndrome Ireland nicola@downsyndrome.ie

Powered by Qualtrics

If you answered 'Other' in the last question please specify your preference.

What do you think is a good session length for you and your child?

- ○ 1 hour
- ○ 40 minutes
- ○ 30 minutes
- ○ 15 minutes

If you were to combine possible options in relation to the two previous questions would your preference be

- ○ 1 hours once a fortnight
- ○ 40 minutes once a week
- ○ 20 minutes 3 times a week
- ○ 10 minutes 3 times a week
- ○ A different combination not specified

If you answered 'A different combination' please specify your

Table 1. *Supplemental* - Vocabulary List and Corresponding Dose for Each Song

Lesson 1					
Baby Shark			The Wheels on the Bus		
Vocabulary Item (n=5)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n=15)	Total Number (per song)	Total Number (per video)
Baby	8	24	Bus	14	42
Mommy	4	12	Day	7	21
Daddy	4	12	Baby	2	6
Granny	4	12	Crying (wa, wa, wa)	4	12
Grandad	4	12	Mommy	2	6
			Daddy	2	12
			Sleep	4	6
			Money	4	12
			Please	4	12
			Doors	2	12
			Open	4	6
			Close	4	12
			People	2	12
			Up	4	6
			Down	4	12

Lesson 2					
Let's Go to the Zoo			Down in the Jungle		
Vocabulary Item (n = 15)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 12)	Total Number (per song)	Total Number (per video)
Go	14	42	Down	5	15
Zoo	14	42	Goes	5	15
Elephant	2	6	Little	5	15
Jump	2	6	Elephant	1	3
Rabbit	2	6	Washing	10	30
Sing	2	6	Clothes	10	30
Monkey	2	6	Here	5	15
Walk	2	6	There	5	15
Chicken	2	6	Lion	1	3
Slide	2	6	Monkey	1	3
Snake	2	6	Bird	1	3
Swim	2	6	Frog	1	3

Fish	2	6
Dance	2	6
Animals	2	6

Lesson 3

This is the Way We			Goodnight Baby Shark		
Vocabulary Item (n = 11)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 7)	Total Number (per song)	Total Number (per video)
Bath	4	12	Bath	4	12
Sleep	7	21	Brush Teeth	4	12
Wash	4	12	Go	4	12
Hair	4	12	Bed	4	12
Dry	4	12	Book	4	12
Nappy	4	12	Read	4	12
Brush Teeth	4	12	Sleep	4	12
Read	4	12			
Book	4	12			
Say	4	12			
Night, night	4	12			

Lesson 4

Let's Go to the Shop			Down by the Sea		
Vocabulary Item (n = 14)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 19)	Total Number (per song)	Total Number (per video)

Go	4	12	Down	4	12
Bus	2	6	Sea	4	12
Food	2	6	Water	1	3
Shop	2	6	Small	1	3
Hungry	2	6	Children	3	9
Stop	2	6	Swim	4	12
Apple	2	6	Here	4	12
There	4	12	There	4	12
Look	1	3	Goes / Go	1	3
See	2	6	Playing	2	6
Sweets	2	6	Wash / Washing	4	12
Me / I	2	6	Clean	1	3
Love	3	9	Eating	2	6
Please	2	6	Ice-Cream	2	6
			Home	2	6
			Mommy	1	3
			Looking	2	6
			Phone	1	3
			Day	1	3

Lesson 5

Can You Roll the Big Ball?			Teddy Bear is Quiet		
Vocabulary Item (n = 8)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 9)	Total Number (per song)	Total Number (per video)
Come	18	54	Teddy Bear	12	36
Roll	26	78	Quiet	12	36
Big	18	54	You	4	12
Ball	18	54	What	4	12
Daddy	2	6	Do	4	12
Mommy	2	6	Jump	5	15
Friends	2	6	Walk	5	15
Me	2	6	Run	5	15
			Clap	5	15

Lesson 6					
Let's Make a Sandwich			Are You Hungry?		
Vocabulary Item (n = 13)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 8)	Total Number (per song)	Total Number (per video)
Make / Do	13	39	You	12	36
Sandwich	13	39	Hungry	4	12
You	3	9	Yes	24	72
Me / I	5	15	Like	8	24
Hungry	2	6	Bananas	2	6
Bread	2	6	Apples	2	6
Want	4	12	Oranges	2	6
Butter	2	6	Grapes	2	6
Egg	2	6			
Chicken	2	6			
Tomato	1	3			
Please	1	3			
Cheese	1	3			

Lesson 7					
Teddy Bear is Sick Today			Can I have?		
Vocabulary Item (n = 15)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 17)	Total Number (per song)	Total Number (per video)
Teddy Bear	6	18	I	3	9
Sick	6	18	Hungry	3	9
Today	6	18	Have	10	30
What	3	9	Food	1	3
Say	3	9	Eat	6	18
Call	4	12	Biscuit	7	21

Doctor	3	9	Box	1	3
Home	1	3	Go	3	9
Good	3	9	Bed	3	9
Get	4	12	Drink	7	21
Medicine	3	9	Little	2	6
Shop	1	3	Milk	2	6
Give	3	9	Juice	3	9
Kiss	3	9	Glass	2	6
Do	1	3	Yes	3	9
			Brush Teeth	3	9
			Night, night	3	9

Lesson 8

What is the Weather?			How's the Weather?		
Vocabulary Item (n = 12)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 11)	Total Number (per song)	Total Number (per video)
What	7	21	Weather	2	6
Weather	7	21	Look	8	24
Today	4	8	Outside	11	33
Sunny	9	27	Sunny	2	6
Raining	2	6	Yes	4	12
Nice	2	6	Play	2	6
Day	3	9	Raining	2	6
Hot	1	3	Inside	1	3
Sea	1	3	Cold	4	12
Go	1	3	Go	2	6
Play	1	3	Snowing	2	6
Me	1	3			

Lesson 9

Baby Shark			The Wheels on the Bus			This is the Way We		
Vocabulary Item (n = 5)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 15)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 11)	Total Number (per song)	Total Number (per video)
Baby	8	16	Bus	14	28	Bath	4	8
Mommy	4	8	Day	7	14	Sleep	7	14
Daddy	4	8	Baby	2	4	Wash	4	8
Granny	4	8	Crying (wa, wa, wa)	4	8	Hair	4	8
Grandad	4	8	Mommy	2	4	Dry	4	8
			Shh	4	8	Nappy	4	8
			Daddy	2	4	Brush Teeth	4	8
			Sleep	4	8	Read	4	8
			Money	4	8	Book	4	8
			Please	4	8	Say	4	8
			Doors	2	4	Night, night	4	8
			Open	4	8			
			Close	4	8			
			People	2	4			
			Up	4	8			
			Down	4	8			

Lesson 10

Down in the Jungle			Let's Go to the Zoo			Let's Go to the Shop		
Vocabulary Item (n = 12)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 15)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 14)	Total Number (per song)	Total Number (per video)
Down	5	10	Go	14	28	Go	4	8
Goes	5	10	Zoo	14	28	Bus	2	4
Little	5	10	Elephant	2	4	Food	2	4
Elephant	1	2	Jump	2	4	Shop	2	4
Washing	10	20	Rabbit	2	4	Hungry	2	4
Clothes	10	20	Sing	2	4	Stop	2	4
Here	5	10	Monkey	2	4	Apple	2	4

There	5	10	Walk	2	4	There	4	8
Lion	1	2	Chicken	2	4	Look	1	2
Monkey	1	2	Slide	2	4	See	2	4
Bird	1	2	Snake	2	4	Sweets	2	4
Frog	1	2	Swim	2	4	Me / I	2	4
			Fish	2	4	Love	3	6
			Dance	2	4	Please	2	4
			Animals	2	4			

Lesson 11

Can You Roll the Big Ball?			Teddy Bear is Quiet			Teddy Bear is Sick Today		
Vocabulary Item (n = 8)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 9)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 15)	Total Number (per song)	Total Number (per video)
Come	18	36	Teddy Bear	12	24	Teddy bear	6	12
Roll	26	52	Quiet	12	24	Sick	6	12
Big	18	36	You	4	8	Today	6	12
Ball	18	36	What	4	8	What	3	6
Daddy	2	4	Do	4	8	Say	3	6
Mommy	2	4	Jump	5	10	Call	4	8
Friends	2	4	Walk	5	10	Doctor	3	6
Me	2	4	Run	5	10	Home	1	2
			Clap	5	10	Good	3	6
						Get	4	8
						Medicine	3	6
						Shop	1	2
						Give	3	6
						Kiss	3	6
						Do	1	2

Lesson 12								
Let's Make a Sandwich			Are You Hungry?			How's the Weather?		
Vocabulary Item (n = 15)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 8)	Total Number (per song)	Total Number (per video)	Vocabulary Item (n = 11)	Total Number (per song)	Total Number (per video)
Make / Do	13	26	You	12	24	Weather	2	4
Sandwich	13	26	Hungry	4	8	Look	8	16
You	3	6	Yes	24	48	Outside	11	22
Me / I	5	10	Like	8	16	Sunny	2	4
Hungry	2	4	Bananas	2	4	Yes	4	8
Bread	2	4	Apples	2	4	Play	2	4
Want	4	8	Oranges	2	4	Raining	2	4
Butter	2	4	Grapes	2	4	Inside	1	2
Egg	2	4				Cold	4	8
Chicken	2	4				Go	2	4
Tomato	1	2				Snowing	2	4
Please	1	2						
Cheese	1	2						

Supplemental Table 2

Post-intervention Questionnaire to establish acceptability
What did you think of this programme?
What would you change about the programme and why?
What were your top three favourite songs and why?
Did the amount of time and energy needed to participate in the programme work for you? (YES /NO please give reasons for your answer)
Did something else in your day /routine / life have to give way so that you could be part of this programme? (YES /NO please give reasons for your answer)
Did the programme fit in with your family values? (YES /NO please give reasons for your answer)

Do you understand the purpose of the programme and how it works? (YES/ NO) please give reasons for your answer

Do you think the programme achieved its purpose? (YES /NO please give reasons for your answer)

Do you feel confident that you can carry on with the programme yourself? (YES /NO please give reasons for your answer)

Is there anything else you'd like to tell us?

Table 3. Supplemental - Exploratory interaction model of dosage with baseline DSE performance

	Understand and Sign			**Imitate**			**Use Spontaneously**		
Predictors	*OR*	*CI*	*p*	*OR*	*CI*	*p*	*OR*	*CI*	*p*
High dose (vs low dose)	0.60	0.31 – 1.14	0.118	0.96	0.55 – 1.68	0.897	1.05	0.60 – 1.81	0.874
Baseline	1.07	1.04 – 1.09	**<0.001**	1.19	1.14 – 1.25	**<0.001**	1.26	1.20 – 1.33	**<0.001**
Group X Baseline interaction	1.09	1.03 – 1.15	**0.004**	1.05	0.96 – 1.14	0.331	0.99	0.91 – 1.08	0.839
Observations	50			50			50		
R^2	0.523			0.518			0.514		
AIC	-164.406			-299.613			-325.521		

Table 3. Supplemental - Exploratory interaction model of dosage with baseline DSE performance

	Understand and Sign			**Imitate**			**Use Spontaneously**		
Predictors	*OR*	*CI*	*p*	*OR*	*CI*	*p*	*OR*	*CI*	*p*
High dose (vs low dose)	0.60	0.31 – 1.14	0.118	0.96	0.55 – 1.68	0.897	1.05	0.60 – 1.81	0.874
Baseline	1.07	1.04 – 1.09	**<0.001**	1.19	1.14 – 1.25	**<0.001**	1.26	1.20 – 1.33	**<0.001**
Group X Baseline interaction	1.09	1.03 – 1.15	**0.004**	1.05	0.96 – 1.14	0.331	0.99	0.91 – 1.08	0.839
Observations	50			50			50		
R^2	0.523			0.518			0.514		
AIC	-164.406			-299.613			-325.521		