Templates and temporality: an investigation of rhythmic motor production in a young man with Down Syndrome and Hearing Impairment

Tracy Jeffery, Sandra P. Whiteside

Abstract

The musical rhythmic abilities of people with Down Syndrome (DS) reportedly exceed their general cognitive abilities. Although they display atypical rhythmic production in musical motor tasks, little is known about how their developmental and cognitive differences affect temporal perception or production. Additionally, hearing impairment often excludes potential participants from studies, further limiting our understanding. This case study examined the rhythmic ability of one young adult with DS and moderate-severe hearing impairment when producing limb-motor movements to music in auditory-only and auditory-visual conditions. His temporal accuracy was observed for gross-motor movements to music. Measurements were made of his temporal accuracy and stability when tapping on a drum at three tempi, and when tapping to non-isochronous rhythms. Results revealed temporal deficits in all tasks. However, production improved in tasks that were augmented with visuo-spatial stimuli, and in isochronous beat entrainment at the fastest tempo. Findings were compared to the participant's cognitive and perceptual profile. Results suggest that his production was limited by developmental factors, but that auditory memory deficits and hearing may account for instability in isochronous entrainment tasks and delayed onset of timing. Recommendations are made for supporting skills in production and perception of auditory rhythms within the DS population.

Keywords

Down Syndrome, rhythm, auditory memory, hearing impairment

Despite a reported sense of musical rhythm in people with Down Syndrome (DS), there has been little systematic investigation of their rhythmic ability in musical tasks (Hooper, Wigram, Carson & Lindsay, 2008; Stephenson, 2006). In part, this may be due to the complexity of individual needs within the DS population. The additional chromosome that causes DS results in a well-documented phenotype, but one that varies in severity between individuals (Silverman, 2007). Anatomical and physiological differences affect physical structures and function, and contribute to motor delays and differences (Latash, 2007; Vicari, 2006), including poor motor coordination (Latash, 2007). Cognitive differences include learning delays (Silverman, 2007), deficits in auditory perception (Roizen, Wolters, Nicol & Blondis, 1993; Pekkonen, Osipova, Sauna-Aho & Arvio, 2007), and poor verbal short term memory (Baddeley & Jarrold, 2007). Auditory processing (AP) deficits may affect the perception of speech sounds, including the rhythm in words and nonsense words (Mason-Apps, Stojanovik & Houston-Price, 2011; Pettinato & Verhoeven, 2009). Additionally, many people with DS are at risk of hearing impairment (HI) (Bennetts & Flynn, 2002; Laws & Hall, 2014).

Although historically described as demonstrating rhythmic abilities in music tasks (Lense & Dykens, 2011), people with DS demonstrate differences in comparison to typically-developing (TD) peers of the same chronological age (CA), and to other groups of similar mental age (MA). Early studies report poorer performance in rhythm discrimination relative to TD controls (Blacketer-Simmonds, 1953), and in rhythm imitation tasks, relative to controls matched for MA (Cantor and Girardeau, 1959). Observational studies highlight inaccurate timing of motor movements to the beat in DS children (Stratford & Ching, 1983; Stratford & Ching, 1989; Picard, 2009) and adults (Picard, 2009). Most studies attribute their inaccuracies in rhythmic timing to motor differences and developmental delay, but one study suggested that rhythmic motor-timing difficulties in DS may be specific to types of motor movements. Stratford and Ching (1989) compared 24 Chinese children with DS (aged 13.2 years) to MA-matched peers when reproducing different whole body and arm rhythms. Both groups were taken from two schools for children with special educational needs. The DS group performed significantly less well than the non-DS group on 'hammering' and 'butterfly' arm-movements. The authors concluded that the DS participants were hampered by motoric limitations in some limb motor-movements, but not all. They also reported a statistically significant correlation between the children with DS and the school setting: those children with DS whose school provided a music curriculum were more accurate than pupils with DS who attended the second school and non-DS participants at both schools. Stratford and Ching (1989) concluded that the DS group had a better capacity to develop their music skills relative to the children with other intellectual impairments.

Two recent studies confirm that DS subjects display atypical motor-movements when striking drums bimanually but demonstrate that the mode of instruction can affect their accuracy (Ringenbach Allen, Chung & Jung., 2006; Chen, Ringenbach, Biwer & Riekena, 2015). In comparison to TD controls matched for CA, Ringenbach and colleagues (2006) reported atypically curvilinear movements in 10 DS adult participants (mean CA 30.2 years; mean MA 7.2 years) and in MA-matched controls when continuously drumming with drumsticks in time with both verbal instructions ("up" and "down") and auditory instructions (the sound of a drum being hit to strike, and a cymbal sound to raise sticks). The DS group and MA-control group also struck the drum out-of phase more often than TD controls. The authors attribute these effects to cognitive delay. However, when shown a video of drumsticks moving in time (visuo-spatial instruction), the movements of the DS and MA control groups better matched those of the TD control group. For the DS group only, the visuo-spatial instruction improved the proportion of in-phase beats. The authors attribute the difference in performance to a deficit in processing verbal instructions which can be mediated by visuo-spatial instruction. Atypical verbal processing was confirmed in a similar study of 14 DS children and adults, in comparison to TD controls (Chen et al., 2015). The study used neuroimaging to measure which area of the brain was dominant when drumming bimanually to verbal, rhythmic, and melodic stimuli at intervals of 500 ms. The results showed a right-hemisphere dominance in the DS participants for processing verbal instructions for rhythm.

Evidence from the bimanual drumming studies (Ringenbach et al, 2006; Chen et al., 2015) suggests benefits for visual modality over auditory-verbal modes for DS participants. In TD populations, visual teaching strategies have led to improvements in rhythmic skills (Sadakata, Hoppe, Brandmeyer, Timmers & Desain, 2008), violin pitch-playing skills (Wang, Wang, Chen, Chang, & Chen, 2012) and singing skills (Howard, Brereton, Welch, Himonides, DeCosta, Williams, & Howard, 2007; Wilson, Lee, Callaghan & Thorpe, 2008), including children with HI (Welch, Saunders, Edwards, Palmer, Himonides, Knight et al., 2015). Such research with people with DS is limited, but visuo-spatial instruction has successfully improved piano skills in individuals with DS (Velasquez, 1991, cited in Peters, 2000). Furthermore, the use of real-time visual feedback with DS children can improve the positioning of speech articulators in words (Wood, 2010). This points to the potential value of visual feedback in supporting musical motor behaviours in DS.

Collectively, research indicates that a range of factors may affect musical motor movement in people with DS. However, group studies make it difficult to isolate causal or contributory factors. Furthermore, previous studies exclude people with DS and identified hearing loss, making it difficult to understand the challenges that are faced by this sub-group. The present case study examines the rhythmic motor behaviours of a young man with DS, Severe Learning Difficulties (SLD) and moderate-severe HI, known here as Robert. His ability to move or tap to isochronous (equal timing between auditory events) and non-isochronous (irregular timing between auditory events) rhythms is measured in response to auditory-only stimuli, and with visuo-spatial support. Robert's abilities in these tasks are compared to his auditory memory and HI and are analysed with reference to previous research on typical and atypical music development. The case study will address the following research questions:

1. How accurately does Robert match motor movements to auditory and auditory-visual stimuli during gross-motor and limb-motor rhythm and beat matching tasks?
2. Given Robert's perceptual and cognitive profile, which factors or combination of factors can best explain changes in his performance within tasks, and between tasks?
3. How might this information be applied to the wider DS population?

Given Robert's developmental delay, he is expected to demonstrate rhythmic errors and characteristics found in younger participants, reflecting his nonverbal MA more closely than his CA. His HI may demonstrate an effect on his motor production. However, it is anticipated that a visuo-spatial stimulus will lead to improved performance by compensating partly or wholly for his HI. Examination of beat entrainment tasks at different tempi may indicate motoric limitations or perceptual limitations associated with hearing or auditory memory capacity. The outcomes may reveal areas of ability or challenge for Robert. This in turn may provide insight into how to support Robert's development, and that of the wider DS population.

Method

A single case study was used to address the research questions, above. The case study is drawn from a larger explanatory series of case studies that focused on speech and musical abilities in four adults with DS, who were resident at a specialist care home (Jeffery, 2016). Explanatory case studies seek to explain links between phenomena that cannot be understood through experimental studies (Yin, 2011). This is appropriate for investigating under-researched or poorly understood aspects of behaviour, and heterogeneous groups such as those with DS and HI.

Ethics

Approval for the study was granted by the Human Communication Sciences' Ethics Committee, University of Sheffield. Informed consent was first sought and obtained from the residential care home. Potential participants were identified by the Principal of the care home using the following inclusion criteria:

1. a diagnosis of DS;
2. aged between 11-25 years;
3. a recognised degree of speech impairment; and
4. an interest in singing.

Staff approached participants known to enjoy music and singing. As the study required participants to perform singing tasks and musical tasks, an interest in singing was required to minimize any potential emotional harm that may be caused by performance anxiety in group sessions. However, it is acknowledged that participants with a self-expressed interest in music may perform differently to peers with DS who do not enjoy performing. Letters and information sheets were provided for staff and parents/carers. Staff distributed the information to parents/carers and provided potential participants with pictorial information sheets that explained the outline of the research. The same staff sought informed consent from the participants after a period of two weeks. The participants retained the right to refuse to participate or to withdraw from assessment tasks as they were presented: consent was therefore ongoing. The participant in this study gave consent to take part in the study and for his data to be used.

Participant

Data were collected from a male adult participant, Robert, who was 23;10 years old, with Severe Learning Difficulties (SLD). Of the four participants in the original study (Jeffery, 2016), Robert was unique in having undergone a recent audiogram. This allowed his musical ability to be examined in consideration of his HI. The audiogram provided by the home's Speech and Language Therapist (SaLT) showed a loss in Robert's right ear of 50-60 dB at 250-1000 Hz and at 4000 Hz, and a loss of 46 dB at 2000 Hz. The loss in his left ear was 40 dB at 250 Hz and 60-65 dB at 500-4000 Hz.

As part of the larger study, Robert had participated in a range of cognitive assessments. The *British Picture Vocabulary Scale (BPVS),* version 1 (Dunn, Dunn, Whetton, & Pintilie, 1992) measures receptive vocabulary, and correlates to verbal intelligence. Although standardised for use for children aged 3-16 years, it has been successfully used with people with DS across a range of abilities (Carr, 2005; Glenn & Cunningham, 2005). The *Goodenough-Harris Draw-a-Person test (DAP)* (Goodenough, 1963) has been used to estimate non-verbal cognitive development in people with learning disabilities. The outcome correlates to visual-motor development and intelligence (Dykens, 1996; Sourtiji, Hosseini, Soleimani & Hosseini, 2010). The *TAPS-R* (Gardner, 1996) task is used as an indicator of verbal short-term memory (standardised for participants of 4-18 years). Participants listen to and repeat a sequence of numbers that are spoken at an even rate of approximately one digit per second, by the examiner. The *Children's Test of Nonword Repetition (CToNWR)* (Gathercole & Baddeley, 1996) measures phonological memory and does not draw on lexical or semantic knowledge. The digits and nonwords were presented live for imitation, with one repetition allowed. The outcomes of these cognitive measures are summarized in Table 1.

Participant profile: hearing abilities and cognition

Robert has a bilateral moderate-severe hearing loss and does not wear a hearing aid. His HI will have consequences for the perception of some speech sounds and frequencies (Vatti, Santurette, Pontoppiddan & Dau, 2014) but will not necessarily affect either perception or enjoyment of music (Matsubara, Terasawa, & Hiraga, 2014).

The data confirm impaired digit span, and deficient performance in verbal and non-verbal intelligence tests relative to his CA of 23 years 10 months. Robert's scores for verbal MA of 3.0 years (2.8 -3.5 years: Table 1) is below the mean MA of 3.79 years reported by Laws and Gunn (2004) when using BPVS to assess 30 children with DS (aged 10-24; mean CA 16.05 years). As BPVS measures receptive vocabulary, it is possible that Robert's relatively low performance on this test may be related to his uncorrected HI which could affect his vocabulary growth; participants in the study by Laws and Gunn (2004) wore hearing aids. Robert's nonverbal MA of 4.25 years as measured by *DAP* (Table 1) is higher than his verbal MA. This is in line with some previous studies that report a higher nonverbal MA (see Naess, Lyster, Hulme & Melby-Lervåg, 2011, for a review). Although *DAP* may be an inappropriate indicator for nonverbal MA in relation to modern psychometric testing (Laws & Lawrence, 2001), it is considered indicative of motor development (Sacks & Buckley, 2003), which is correlated with MA in people with DS (Chen et al., 2015; Sourtiji et al., 2010).

Robert was unable to reproduce nonwords in the *CToNWR* task, a measure of phonological memory (Table 1). Robert's *TAPS* score (Table 1) was 2 digits. It is difficult to isolate the effects of reduced short-term memory (STM) from his HI, but during the TAPS task Robert verbally repeated numbers (e.g. the word *three*) in two-digit sequences that he failed to reproduce in sequences of three digits. This indicates that the test did successfully measure his memory, rather than his perception of words. A mean digit span of 3-4 digits is typically reported in people with DS (Bird, Cleave &

McDonnell, 2000; Seung & Chapman, 2000) irrespective of the CA of participants (Purser & Jarrold, 2005).

Table 1: Cognitive measures
The table shows the cognitive measures that the participant undertook, the tests used, the scoring system, and the results

Cognitive measure	Test	Measure	Result
Verbal Mental-Age equivalent	British Picture Vocabulary Scale (BPVS) 1	Raw score	25
	BPVS 1	Age equivalent (years; months) and range (years; months)	3;0 m (2;8 - 3;5)
Non-Verbal Mental-Age equivalent	Draw-A-Person (DAP)	Age equivalent (years; months)	4; 3
Auditory memory	Digit Span	Max. number of digits correctly imitated	2
Phonological memory	Children's Test of Nonword Repetition (CToNWR)	Number of nonwords correctly imitated	0/10

Measures

Temporal measures

Three tasks were developed and administered to assess motor-musical ability in limb movements: Gross motor skills to music; Beat entrainment; Rhythm entrainment. Details are given below.

Gross motor skills to music

Gross motor movements were assessed visually against a checklist during live performance. The checklist (Table 2) had been developed by the first author for teaching groups of comparable abilities. Robert was asked to perform a selection of limb-motor movements in time to the beat of *Beat It*, by Michael Jackson, which has a median tempo (138 beats per minute (bpm): www.bpmdatabase.com) and was familiar to Robert.

Beat entrainment

This task assessed the ability to synchronise to a beat at different tempi. The task is based on that of Peter and Stoel-Gammon (2008) who examined entrainment in relation to developmental apraxia of speech. This test used the same tempi of 104, 132, and 160 bpm. A simple metronome sequence was created on *Garageband* 08 Version 4.1.2 (Apple inc. 2002-2007) using a 'clave' sound from within the *Garageband* loop section. The loop was edited to place the 'strong' beat in first position, and the second beat of each sequence was copied and placed on beats 2, 3 and 4. The new loop was copied to create a metronome with a strongly metrical beat. Robert was asked to listen for four bars

(16 beats), then tap on a small hand-drum in time to the beat for five bars (30 beats) and to maintain the beat when the sound stopped.

Rhythm entrainment

A clave sample was selected from the *Garageband* loops, that consisted of binary rhythms consisting of half and quarter notes (*ta tee-tee ta ta*). This sample was looped and aligned to the first beat of each bar to create a simple 10-bar pattern. Robert was instructed to listen to the first two bars, and to then join in on the drum after a verbal cue of *1,2,3, go*.

Procedures

Assessments

Assessments were presented via an *iMac*, and amplified through an *Amethyst iPig* speaker, adjusted to a volume level that was comfortable for Robert. Pictures or Makaton were used during assessments to support his comprehension of the tasks. Assessments took place during a single 2-hour afternoon session in a private room adjacent to the care home's dining room. There was intermittent noise from the adjacent room during assessments.

Recordings

Audio recordings were made using a shock-mounted cardioid condenser *RODE NT1A* microphone that was connected to the computer via a soundcard. Responses were recorded directly into *Garageband*.

Gross motor skills

Judgements were made based on observation of Robert's live performance. A rating scale (Table 2) was devised to indicate the observed accuracy of the movements relative to the beat. Robert was asked to imitate movements initially, and an impressionistic score was given. For each movement, the first researcher then produced the desired movements as ongoing visuo-spatial support and an impressionistic score was given.

Analysis of beat and rhythm entrainment

Recordings of beat and rhythm entrainment tasks were imported into *Praat* vs. 5.4.05 (Boersma & Weenink, 2011), which enables precise measurement of duration between successive acoustic features. The audio recording, waveform and intensity peaks were used to determine the onset of target beats. These were marked as 'boundary lines' on the first tier of the text grid (Figure 1). Robert's taps were identified in the same manner and were marked as boundary lines on the second tier of the text grid. This enabled measurement of the timing between the stimulus (Tier 1) and Robert's productions (Tier 2), generating a value, in milliseconds, for *Inter-tap Interval* (ITI). The values between successive taps were annotated on the text grids in the space between each boundary line. A similar method enabled measurement of the *Anticipation Time* (AT) for beat entrainment tasks. Selection of the duration between the participant’s timing, on Tier 2, and the target beat (Tier 1) enabled measurement of AT, which was recorded on Tier 3.

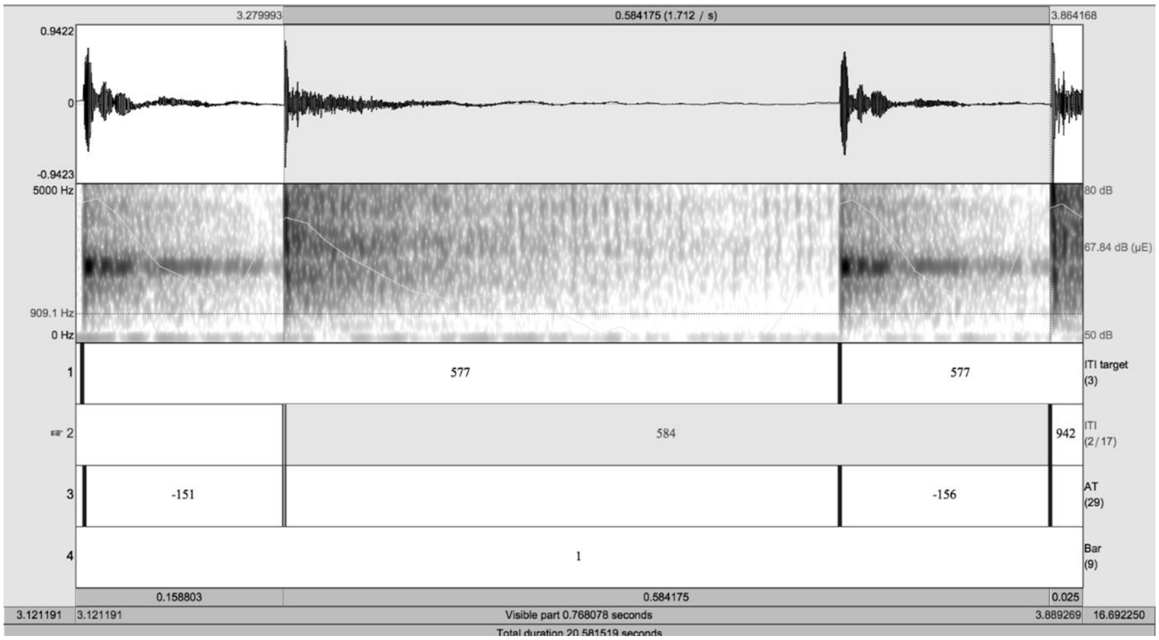

Figure 1: Calculation of the Beat Entrainment data using Praat. The text grid shows the waveform (top) and spectrogram of an extract of the beat entrainment task at 104 bpm. The duration between successive taps (ITI) in the target beat is marked in Tier 1, in milliseconds. The ITI of Robert’s taps is marked in Tier 2, and the Anticipation Time (AT, ms) in Tier 3. Robert’s taps were produced after the target beat, resulting in a negative AT. The Bar number is shown in Tier 4.

For beat entrainment scores, descriptive statistics were generated in SPSS v21 (IBM Corp, 2012). For rhythm, the ITI scores were input into Pages v5.6.1 (Apple Inc., 2015), and a graph was generated to illustrate the duration of successive ITI values. A moving average was also generated, as an indicator of the similarity in durational rhythmic pattern between the target and Robert’s production. As Robert’s reproduction did not appear to align with the target at any point, AT scores were not generated.

Reliability

After a period of 12 months, data from beat entrainment tasks were re-measured by the first author, from the original recordings. All measures at 104 bpm (20 taps) were re-measured, whose results were the most variable; and 25% of measures at 132 bpm and 160 bpm. These were taken from the onset of playing that was at the tempo most appropriate for the task: that is, if a visual cue was given, the measurement was taken from the participant’s first tap. A Pearson’s correlation test was applied in SPSS v21. The results indicated high intra-rater reliability, with statistically significant ($<0.05^*$) effects for AT at 160 bpm ($r = .869$). Correlation was highly-significant ($p<0.01^{**}$) for AT and ITI at 104 bpm ($r = .850$; $r = .997$); AT and ITI at 132 bpm ($r = .932$; $r = .999$); and for ITI at 160 bpm ($r = 0.845$).

Results

Gross motor-movements

Robert had difficulties in coordinating movements to the beat in aural and visually-supported conditions (Table 2). When given visual support that continuously modelled the desired movements and the timing of these to the dominant hierarchical beat, Robert's movements were typically slower than the demonstration and lagged behind the beat. Robert seemed to have difficulty in alternating leg movements and moved just his left leg in response to the beat. When asked to walk in time to the music, he moved slowly and watched his own feet as he did so. He was unable to alternate his arms when asked to lift one at a time from his side: instead he moved his upper torso in the indicated direction.

Table 2. Gross motor movement scores in aural and audio-visual conditions. The table shows the scores allocated to Robert when reproducing (aural condition) or imitating continuous (visual assistance) gross motor movements to the song *Beat It* by Michael Jackson. The criteria and range for scores are given at the foot of the table.

Target motor movement, in time to the pulse	Aural condition only	Aural with ongoing visual assistance
Tap dominant foot	2	2
Step on the spot, alternating feet	0	0
Walk around the room	1	2
Lift both arms out to the side, and drop	1	2
Lift alternate arms out to the side, and drop	0	1
Clap hands	2	2
Raise both shoulders towards ears	2	2
Raise alternate shoulders towards ears	2	2
Twist torso	2	3
Independently, create a movement	2	n/a
Score: total/maximum score (%)	14/50 = 28%	16/45 =35.55%

Scoring:

0	1	2	3	4	5
no participation/unable to perform task	no regularity: wide variation between successive beats	maintains a regular tempo, but at own rhythm, not imposed	movements are mostly regular and are sometimes in time eg. for short time or with visual assistance. But lag behind/in front	movements are regular and coincide with most strong beats	accurate: feet coincide with beat

Beat entrainment

Data from the beat entrainment tasks are given in Table 3. Robert adapted his tempo (ITI, ms) in response to changes in the tempi of the beat entrainment tasks. The duration between his beats was variable (SD of ITI, %). The pace and regularity of his movements (ITI, ms, and SD of ITI, ms respectively) were most accurate at 160 bpm. In all tasks, Robert was unable to synchronise to the beat (AT, ms) and his timing was inconsistent (SD of AT, ms). His timing to the beat was most accurate (AT accuracy, %) and least variable (SD of AT, ms) at 160 bpm. However, at 160 bpm, he played close to anti-phase (53 % AT).

Table 3: Beat entrainment. Mean results for tapping to a metronome at tempos of 104 bpm, 132 bpm, and 160 bpm. The table shows the duration of mean Inter-Tap-Interval (ITI) and Anticipation Time (AT) in milliseconds, as a percentage of the target interval, and the SD in milliseconds. The + symbol indicates that Robert's taps occurred after target beats.

Measure	104 bpm	132 bpm	160 bpm
Target Inter-Tap-Interval (ITI), ms	576	455	375
No. of taps produced	20	25	33
ITI, ms	799	583	390
ITI, accuracy %	72	78	96
SD of ITI, ms	145	36	57
Mean range of ITI, accuracy =16%			
AT, ms	+184	+108	+199
AT, accuracy %	32	24	53
SD of AT, ms	191	159	73
Mean AT, accuracy =36%			

Rhythm entrainment

Robert was unable to reproduce the target rhythm or to match taps to the target beat. Figures 2 and 3 show the approximate alignment and approximate duration of his beats to target beats in medial bars of Rhythm 1 (aural only) and Rhythm 2 (aural with ongoing visual support), respectively. Although Robert's pattern does not synchronize with the target and comprises intervals of longer duration, the timeline indicates an attempt to produce some variation in the rhythmic pattern, especially in Rhythm 2 (Figure 3).

Robert's beat, ms			**1**		**2**		**3**		**4**		**5**	**6**		**7**		**8**		**9**		**10**
Target beat, ms	**1**		**2**	**3**	**4**		**5**		**6**		**7**	**8**	**9**		**10**					
Time, ms	0	280	560	840	1120	1500	1780	2060	2340	2620	2900	3180	3460	3740	4020	4300	4580	4860	5140	5420
Bar	4										5									

Figure 2. A timeline of the final 2 bars of Rhythm 1: *Ta tee-tee Ta Ta*. The timeline shows the approximate alignment of Robert's final ten beats in the last two bars of Rhythm 1 relative to the onset of ten Target beats, and the approximate duration (ms) of Robert's beats 1-9. The timeline is divided into equally-spaced units of 280 ms (line 3), which is the average duration of the quarter note (tee). Beat numbers are placed at approximate positions relative to sub-divisions of 140 ms. Robert's beats are longer in duration than target beats, and lag behind the stimulus. Robert was asked to stop playing at his Beat 10 and to 'watch my fingers'. Shaded areas exclude previous beats played by Robert, and continuing target beats for ease of comparison to the Target and to Figure 4.

Robert's beat, ms		**1**	**2**		**3**		**4**			**5**			**6**		**7**	**8**			**9**		**10**
Target beat, ms	**1**	**2**	**3**	**4**	**5**		**6**		**7**	**8**	**9**	**10**									
Time, ms	0	280	560	840	1120	1500	1780	2060	2340	2620	2900	3180	3460	3740	4020	4300	4580	4860	5140	5420	5520
Bar	3								4								5				

Figure 3. A timeline of bars 3-5 of Rhythm 2: *tee-tee tee-tee Ta Ta*. The timeline shows the approximate alignment of Robert's final ten beats in bars 3-5 of Rhythm 2 relative to the onset of ten Target beats, and the approximate duration (ms) of Robert's beats 1-9. The timeline (line 3) is divided into 280 ms, which is the average duration of the quarter note (tee). Beat numbers are placed at approximate positions, relative to sub-divisions of 140 ms. Robert's beats lag behind the stimulus. Although the duration of his beats is greater than the duration of target beats, the duration between beats 1-2 and 7-8 relative to his longer beats reflects the target 'tee-tee' pattern. Shaded areas exclude previous beats played by Robert played in Bar 3 and exclude continuing target beats for ease of comparison to the target and to Figure 5. The task was concluded after the fifth target bar.

The data for Robert's ITI (ms) are given in Figures 4 and 5. These show a difference in Robert's timing between beats when the stimulus was aural only (Figure 4) and when the aural stimulus was accompanied with an ongoing visual demonstration of the rhythm (Figure 5). Although the two rhythms are different in pattern, both patterns consisted of simple subdivisions (the quarter note, *ta*, and eighth-notes, *tee-tee*) and the tempo remained constant at 104 bpm. Robert's ITI in the aural condition varied between 481 ms (Figure 4: beat 1) and 637 ms (Figure 4: beat 10). His range was 156 ms, half that of the stimulus (265-585 ms). The duration between one of his beats was close to the target ITI (Figure 4: beat 10). In the visually-mediated condition (Figure 5), Robert produced a wider range of ITI (255-854 ms), exceeding the stimulus range (261-561 ms). With ongoing visuo-spatial support, the durations in five of Robert's beats were close to the target ITI (Figure 5: beats 1, 5, 6, 7 and 11). In comparison to the aural-only production (Figure 4), Robert's moving average (solid line) is more characteristic of the demonstration (dotted line in the rhythm that included visual demonstration (Figure 5).

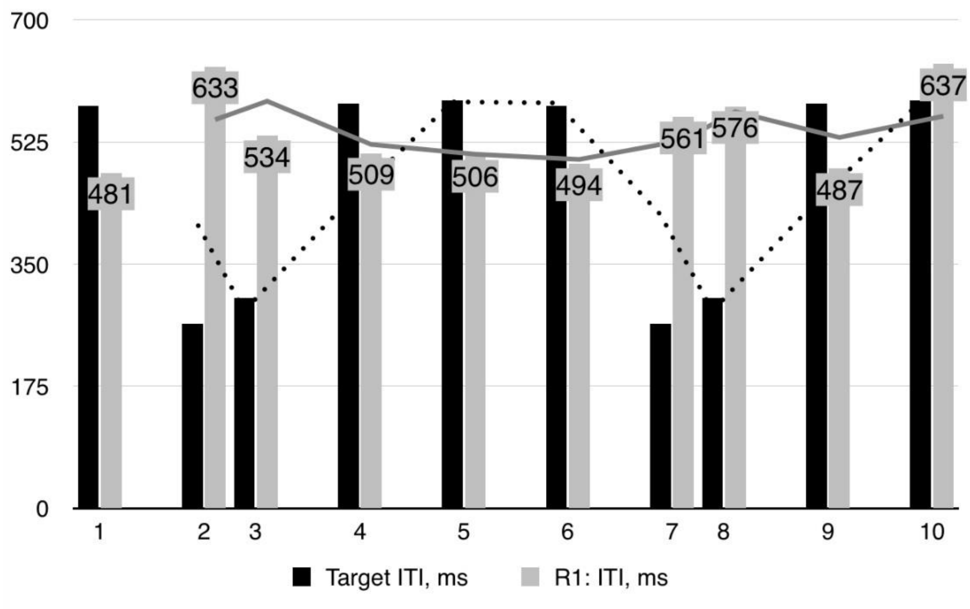

Figure 4: Graph showing the target ITI and the ITI of Robert's production in the Rhythm entrainment task - ta tee-tee ta ta. The chart shows the duration in milliseconds (ITI, ms: vertical axis) between beats (horizontal axis) of the task shown in Figure 2. The black bars show the target ITI (ms), which consists of a pattern of ten beats, in a 2:1 ratio. The grey bars represent the duration between Robert's beats, which were produced in response to the auditory stimulus only. The duration between his beats (ms) is marked at the top of each column. The solid grey and dotted lines show the 'moving average' of Robert's production and of the target rhythm, respectively.

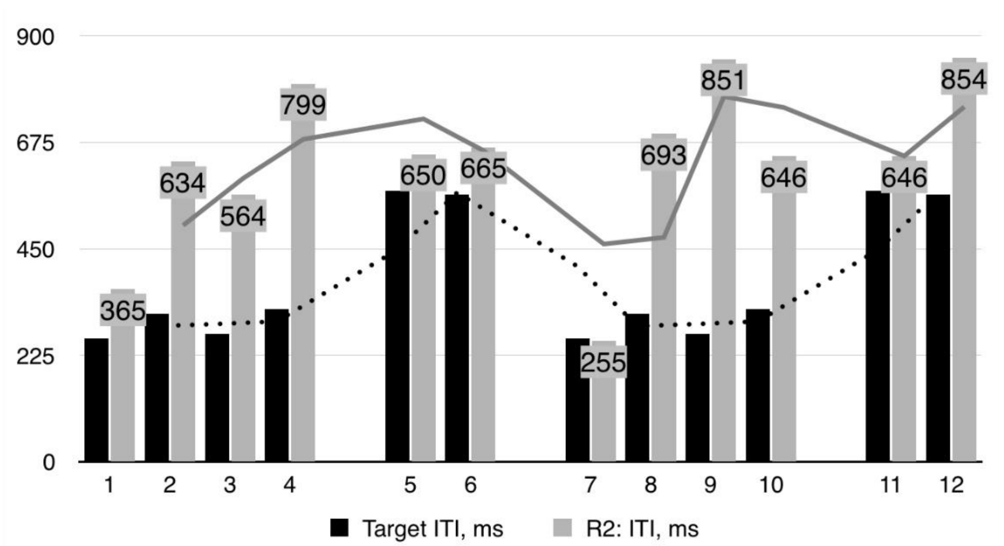

Figure 5: Graph showing the target ITI and the ITI of Robert's production in the second Rhythm entrainment task - tee-tee tee-tee ta ta. The chart shows the duration in milliseconds (ITI, ms: vertical axis) between beats (horizontal axis) of Rhythm 2 (see also Figure 3). The black bars show the target ITI (ms), which consists of a pattern of ten beats, in a 2:1 ratio. The grey bars represent the duration between Robert's beats, which were produced with ongoing visuo-spatial support. The duration between his beats (ms) is marked at the top of each column. The solid grey and dotted lines show the 'moving average' of Robert's production and of the target rhythm, respectively.

Discussion

Observations of Robert's rhythmic gross-motor movements (Table 2) indicate poor co-ordination, slow movements and unstable posture. These observations are consistent with research involving DS participants (Latash, 2007; Rigoldi, Galli, Mainardi, Crivellini & Albertini, 2011; Shumway-Cook & Woollacott, 1985) and suggest a generalized motor difference, as is observed in DS subjects (Sacks & Buckley, 2003; Silverman, 2007). However, HI impacts upon postural balance, speed of motor movements and upon motor co-ordination (for a review, see Rajendram, Roy & Jeevanantham, 2012). The effects of HI on gross-motor abilities in DS individuals are unknown: those with HI were excluded from studies investigating postural control in DS (Shumway-Cook & Woollacott, 1985; Rigoldi et al., 2011) and were not assessed in a study of balance and motor skills (Malak, Kotwicka, Krawczyk-Wasielweska, Mojs & Samborski, 2013). The findings suggest that for Robert, any difficulties in gross-motor functioning and balance that are associated with DS may be exacerbated by his HI.

Robert's performance in rhythmic tapping tasks is consistent with available research in DS that indicates a qualitative difference in rhythmic ability relative to TD controls of equivalent MA (Stratford & Ching, 1989) and age-matched TD controls (Ringenbach et al., 2006). Robert was unable to reproduce isochronous hand movements (Table 2), or non-isochronous hand movements in rhythm entrainment tasks (Figures 2, 3, 4 and 5). This is unlikely to result directly or solely from his HI as he responded independently to tempo changes in the beat entrainment tasks (Table 3) indicating that he heard the stimuli. Therefore, it is necessary to also consider maturational factors, his auditory memory, and how his HI may affect his processing of sound and subsequent production of movements. Each factor is discussed in turn, below.

In beat entrainment, Robert's scores in SD of AT and ITI (%) (Table 3) highlight poor synchronisation to the beat, and variable timing between beats, respectively. In tapping tasks, synchronisation usually develops at about 6-7 years, and stability develops at about 8 years, but Robert's variability exceeds the reported norms (Repp, 2005; Repp & Su, 2013). As accuracy and stability of timing in tapping tasks is linked to motor development (Kuhlman & Schweinhart, 2000; Phillips-Silver, 2009), Robert's inaccuracy could reflect a general motoric delay. Indeed, Robert was more accurate and consistent in his timing between taps (SD of AT, ITI %: Table 3) at faster tempi than at slow tempi, a characteristic typical of younger children (Drake, Jones & Baruch, 2000; Reifinger, 2006; Repp, 2005; Repp & Doggett, 2007). Children of 4 years are more accurate in tapping tasks when the ITI is between 300-400 ms (Drake et al., 2000). Robert played most accurately at 160 bpm (Table 3: 96% accuracy of ITI; close to anti-phase at 53% AT), for which the target ITI is 375 ms. Robert's ability in the task corresponds with his nonverbal MA of 4.3 years (Table 1), indicating a developmental delay, broadly in line with younger TD children. Although a motor delay is possible, an immature ability to attend to or perceive slower rhythmic patterns might also explain this. Drake et al. (2000) explain that immature cognitive factors may cause younger children to fail to attend to or discriminate tempi beyond these limits of 300-400 ms. Auditory memory is essential for reproducing and synchronising to auditory rhythmic patterns (Flaugnacco, Lopez, Terribili, Zoia, Buda, Tilli et al. 2014; Grahn & Schuit, 2012; Saito, 2001. In TD adults, auditory memory lasts 1.5-4 seconds (Grahn & Schuit, 2012). At 160 bpm, one bar is 1.5 seconds in duration, but the duration of a bar at 104 bpm is 2.3 seconds. A digit span of 2 may mean that Robert is unable to perceive and lay down a stable temporal template (Thaut, 2008) at slower tempi, affecting his motor timing and stability more than at faster tempi.

Robert's auditory memory may also explain his temporal production in non-isochronous tapping tasks. He showed little temporal variation in these tasks, which could indicate motoric limitations or hearing limitations. However, patterns of five (Figure 2) and six (Figure 3) notes are likely to have exceeded his 2-digit auditory-verbal memory capacity. With ongoing visual support his movements became increasingly varied, in line with the target rhythm (Figure 3, Figure 5). Visual instruction assists motor performance of DS participants in manual rhythm tasks (Chen et al., 2015; Ringenbach et al., 2006), and for Robert, improved performance with visual support negates motoric differences as a primary limiting factor.

Robert's productions in rhythm entrainment tasks were poorly aligned to the stimulus (Figures 2 and 3) and followed the stimulus (negative AT). Rhythm entrainment does not typically develop until 8-10 years (Drake et al., 2000), so this may indicate a developmental motor delay, given Robert's MA of 4;3 years (Table 1). However, negative AT occurred in Robert's beat entrainment at all tempi (Table 3), which is atypical of younger children who have immature motoric and perceptual abilities (Thaut, 2008). Thaut (2008) attributes such behavior to disturbances to the auditory and proprioceptive feedback that is required to correct timing for the following tap. Robert's improved performance on rhythm tasks when given visual support (Figure 3, Figure 5) also suggests difficulty in linking the auditory signal to motor movement. Differences in how the brain responds to sound have been identified in people with DS (Groen, Alku & Bishop., 2008; Pekkonen et al., 2007) and slow processing time for auditory signals has been demonstrated in DS children (Groen et al., 2008; Porter, Grantham, Ashmead & Tharpe, 2014). Accuracy in tapping tasks requires integration of auditory signals with proprioception and motor output (Repp, 2006; Thaut, 2008). Even in hearing adolescents, reduced perception of the auditory signal has been linked to variable rates in tapping tasks (Tierney & Kraus, 2013). Any processing difficulties in DS will be exacerbated by bilateral hearing loss (Marcell, Cohen, Weathers, Wiseman, Croen & Sewell, 1990; Marcell, 1995). For Robert, AP deficits and HI may combine to affect his timing.

In conclusion, it is not possible to rule out motoric deficits when considering the timing deficits displayed by Robert, as these are a known feature of DS (Latash, 2007; Rigoldi et al., 2011). However, the data indicate that a combination of cognitive (memory), physiological (hearing) and perceptual (processing) deficits contribute to Robert's difficulties. The data also indicate that the use of visual teaching strategies may be particularly useful in developing accuracy in his musical motor movements.

Wider Implications and Recommendations

Previous observational studies of children and adults with DS noted a general ability to make whole body movements in time to recorded music, but reduced accuracy in comparison to TD populations of the same CA (Blacketer & Simmonds, 1953; Stratford & Ching, 1989). According to Stratford and Ching (1989), the accuracy of motor timing movements in DS may depend on developmental limitations, the type of movement, and the duration and type of training. Like many people with DS (Rigoldi et al., 2011; Shumway-Cook & Woollacott, 1985), Robert's gross-motor movements were unstable and poorly co-ordinated. In the TD population, there is evidence that how adults move in time to music has direct consequences for their perception of the beat (Phillips-Silver & Trainor, 2005, 2008). Movement activities involving the vestibular system may therefore be critical in developing a sense of beat and rhythm, and such activities will be especially important for those at risk of HI. For people with DS, developing whole body movements to music, especially when young, may support developing musical perception.

The case study also discussed how Robert's accuracy in motor timing may be affected by perceptual difficulties. Robert's HI is likely to exacerbate his motor coordination deficits, but his digit span may affect his ability to form a mental representation or template. For people with DS, visuo-spatial cues during teaching may help to compensate for deficits in both hearing and auditory-verbal memory. Indeed, people with DS develop greater accuracy in rhythmic motor movements when supported by visuo-spatial cues (Ringenbach et al., 2006). Rhythmic musical training may also improve AP, with relevance to both musical aptitude and speech perception. Although the present study did not measure AP in Robert, his inconsistent ITI and negative AT in beat entrainment tasks suggest poor coupling between auditory information and motor response (Thaut, 2008). Auditory processing may be slow in people with DS (Marcell & Cohen, 1992; Marcell, 1995), but AP responses can develop in response to rhythmic training in TD children (e.g. Kraus & Chandrasekaran, 2010; Parbery-Clark, Skoe, Lam & Kraus, 2009; Parbery-Clark, Anderson, Hittner & Kraus, 2012; Tierney & Kraus, 2013). Tierney and Kraus (2013) argue that practice in beat timing may improve the brain's response to processing auditory signals. There is also increasing evidence that musical training leads to superior AP in those with HI (Matsubara, Terasawa & Hiraga, 2014; Parbery-Clark, Anderson & Kraus, 2013; Rochette, Moussard & Bigand, 2014). For example, in people with HI, musical experience enhances rhythmic tapping performance in adults (Matsubara, Terasawa & Hiraga, 2014). Therefore, there is the potential of rhythmic music training to support and develop musical aptitude in people with DS. Potentially, this may even support language processing and production, as observed in TD language-impaired populations (see Schön & Tillman, 2015, for a review) and HI children, (Cason, Hidalgo, Isoard, Roman & Schön, 2015; Hidalgo, Falk & Schön, 2017).

Research in TD populations shows that engagement in musical activities offers benefits for music production and AP, even for participants with HI. Therefore, it is recommended that music education with DS populations should begin in the early years, when children with DS are at greatest risk of intermittent HI. A range of movement activities may be needed to support the developing vestibular system and improve perception of beat, helping compensate for HI. Visuo-spatial teaching methods will reduce reliance on auditory STM and support perception of target patterns. Activities also need to offer immediate visual, spatial or tactile feedback to limit AP deficits or HI. The use of technologyto provide visual feedback may be particularly successful with the DS group, in line with previous studies with TD populations. Specifically, visual feedback should be used to provide information to the performer about their performance, enabling 'knowledge of results' (Welch, 1998; Welch, Howard & Rush, 1989), especially in those with HI (Welch et al., 2015). Although the present study relied on gestural and simple visual forms of support and feedback, the availability and accessibility of mobile technology make this a useful tool for supporting accuracy in musical skills (e.g. Lin, Anderson, Hamzeen & Lui, 2014). For a population whose majority derive so much enjoyment from music, visually-mediated musical activities offer immediate benefits for developing musical skills and abilities; and for those with HI and AP deficits, the benefits may extend further still.

Conclusion

This study adds to the previous studies of DS participants that indicate a difficulty in accurately timing limb motor-movements to musical stimuli, and that suggest that poor timing may be in line with cognitive development. However, the study also suggests that HI may affect production at the AP level with consequences for motor behaviour; and that a limited digit span may affect the ability to form a mental representation of temporal patterns. The impact of digit span and AP deficits have been identified in the wider DS population in the speech domain but have not been previously studied in relation to musical activities, to the authors' knowledge. Likewise, no known previous study has examined rhythmic abilities in DS participants with significant HI.

Much of the existing data on beat tapping skills relies upon finger-tapping tasks (Patel, 2008). The participant in this study tapped on a drum, which may require a different range of motor movements. This may limit comparison to other studies. Additionally, the data were collected in the participant's daycare environment, which included noise from adjacent living areas. Although the naturalistic setting has ecological validity, excessive noise can contribute to perceptual difficulties and overload phonological working memory (Schellenberg & Weiss, 2012). Further studies would benefit from using tapping tasks in quiet settings. Additionally, a computerized programme using finger taps would enable collection of lengthier samples and automatic calculations, enabling greater cross-comparison and a larger sample-size.

The study did not include repeated assessment of tasks. Given the participant's age and previous experience in music-related performances (Table 1), it is assumed that his performance in music-motor tasks reflects well-established behaviours. Future studies would benefit from a longitudinal design involving younger participants and repeated assessments. This would also allow the effects of intervention to be measured, which in turn could clarify the role of different parameters for people with DS and HI and would indicate more clearly their potential to learn from different strategies.

Although a single case study produces limited data and limits generalization, the approach allows the complex interactions between HI and other factors to be considered. To isolate the role of each parameter, further studies should investigate music timing in beat entrainment tasks in relation to hearing abilities, auditory perception, auditory processing abilities, digit span, and motor development. Such study would further our understanding of how each factor affects temporal production and clarify how best to support musical development within the DS population.

Acknowledgements
The authors wish to thank the participant of the study and the staff at the care home for enabling it.

References

Baddeley, A., & Jarrold, C. (2007). Working memory and Down syndrome. *Journal of Intellectual Disability Research*, 51(12), 925-931.

Bennetts, L. K., & Flynn, M. C. (2002). Improving the classroom listening skills of children with Down syndrome by using sound-field amplification. *Down's Syndrome, Research and Practice*, *8*(1), 19–24. http://doi.org/10.3104/reports.124

Bird, E. K. R., Cleave, P. L., & McConnell, L. (2000). Reading and phonological awareness in children with Down syndrome: A longitudinal study. *American Journal of Speech-Language Pathology*, 9(4), 319-330

Blacketer-Simmonds, D. A. (1953). An investigation into the supposed differences existing between mongols and other mentally defective subjects with regard to certain psychological traits. *The British Journal of Psychiatry*, 99(417), 702-719.

Boersma, P., & Weenink, D. (2011). Praat: Doing phonetics by computer [Computer program]. Version 5.3. 03. 2011.

Cantor, G. N., & Girardeau, F. L. (1959). Rhythmic discrimination ability in mongoloid and normal children. *American journal of mental deficiency*, *63*(4), 621-625.

Carr, J. (2005). Stability and change in cognitive ability over the life span: a comparison of populations with and without Down's syndrome. *Journal of Intellectual Disability Research*, 49(12), 915-928.

Cason, N., Hidalgo, C., Isoard, F., Roman, S., & Schön, D. (2015). Rhythmic priming enhances speech production abilities: Evidence from prelingually deaf children. *Neuropsychology*, 29(1), 102.

Chen C-C , Ringenbach S.D.R., Biwer A., & Riekena, A. A cerebral lateralization of the EEG during perceptual-motor integration in young adults with Down syndrome: A descriptive study. *Brazilian Journal of Motor Behavior*. 2015: 9(2): 1-7.

Drake, C., Jones, M. R., & Baruch, C. (2000). The development of rhythmic attending in auditory sequences: attunement, referent period, focal attending. *Cognition*, 77(3), 251–88. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/11018511

Dunn, L. M., & Dunn, L. M. Whetton, C., & Pintilie, D. (1982). British Picture Vocabulary Scale. Windsor, England: NFER-Nelson.

Dykens, E. (1996). The draw-a-person task in persons with mental retardation: What does it measure? *Research in developmental disabilities*, 17(1), 1-13.

Flaugnacco, E., Lopez, L., Terribili, C., Zoia, S., Buda, S., Tilli, S., ... & Schön, D. (2014). Rhythm perception and production predict reading abilities in developmental dyslexia. *Frontiers in Human Neuroscience*, 8(392), 10-3389.

Gardner, M. (1996). TAPS-R : Test of Auditory-Perceptual Skills (non-motor)-Revised : manual. Psychological and Educational Publications: Hydesville, CA

Gathercole, S. E., & Baddeley, A. D. (1996). Children's Test of Nonword Repetition. Psychological Corporation: London, UK.

Glenn, S., & Cunningham, C. (2005). Performance of young people with Down syndrome on the Leiter- R and British picture vocabulary scales. *Journal of Intellectual Disability Research*, 49(4), 239-244.

Goodenough, F. L. (1963). Goodenough-Harris drawing test. New York: Harcourt Brace Jovanovich.

Grahn, J. A., & Schuit, D. (2012). Individual differences in rhythmic ability: Behavioral and neuroimaging investigations. *Psychomusicology: Music, Mind, and Brain*, 22(2), 105.

Groen, M. A., Alku, P., & Bishop, D. V. M. (2008). Lateralisation of auditory processing in Down syndrome: A study of T-complex peaks Ta and Tb. *Biological Psychology*, *79*(2), 148–157. http://doi.org/10.1016/j.biopsycho.2008.04.003

Hidalgo, C., Falk, S., & Schön, D. (2017). Speak on time! Effects of a musical rhythmic training on children with hearing loss. *Hearing research*, *351*, 11-18.

Hooper, J., Wigram, T., Carson, D., & Lindsay, B. (2008). A review of the music and intellectual disability literature. *Music Therapy Perspectives*, *26*, 66–79.

Howard, D. M., Brereton, J., Welch, G. F., Himonides, E., DeCosta, M., Williams, J., & Howard, A. W. (2007). Are real-time displays of benefit in the singing studio? An exploratory study. *Journal of Voice*, *21*(1), 20-34.

Jeffery, T. (2016). *Speaking in harmony: an exploration of the potential for rhythm and song to support speech production in four young adults with Down Syndrome* (Doctoral dissertation, University of Sheffield).

Kraus, N., & Chandrasekaran, B. (2010). Music training for the development of auditory skills. *Nature Reviews. Neuroscience*, *11*(8), 599–605. http://doi.org/10.1038/nrn2882

Kuhlman, K., & Schweinhart, L. J. (1999). Timing in Child Development. *High/Scope Educational Research Foundation*, 1–10. Retrieved from: http://www.highscope.org/Content.asp?ContentId=234

Latash, M. L. (2007). Learning motor synergies by persons with Down syndrome. *Journal of Intellectual Disability Research*, 51(12), 962–971. http://doi.org/10.1111/j.1365-2788.2007.01008.x

Laws, G., & Gunn, D. (2004). Phonological memory as a predictor of language comprehension in Down syndrome: a five- year follow- up study. *Journal of Child Psychology and Psychiatry*, *45*(2), 326-337.

Laws, G., & Hall, A. (2014). Early hearing loss and language abilities in children with Down syndrome. *International Journal of Language & Communication Disorders*, 49(3), 333-342.

Laws, G., & Lawrence, L. (2001). Spatial representation in the drawings of children with Down's syndrome and its relationship to language and motor development: A preliminary investigation. *British Journal of Developmental Psychology*, *19*(3), 453-473.

Lense, M. D., & Dykens, E. M. (2011). Musical interests and abilities in individuals with developmental disabilities. *International review of research in developmental disabilities*, 41, 265-312.

Lin, K. W. E., Anderson, H., Hamzeen, M. H. M., & Lui, S. (2014). Implementation and Evaluation of Real-Time Interactive User Interface Design in Self-learning Singing Pitch Training Apps. In *ICMC*.

Malak, R., Kotwicka, M., Krawczyk-Wasielewska, A., Mojs, E., & Samborski, W. (2013). Motor skills, cognitive development and balance functions of children with Down syndrome. *Annals of Agricultural and Environmental Medicine : AAEM*, *20*(4), 803–6. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/24364457

Marcell, M. (1995). Relationships between hearing and auditory cognition in Down syndrome youth. *Down Syndrome Research and Practice*, *3*(3), 75–91. http://doi.org/10.3104/reports.54

Marcell, M.M., Cohen, P.S., Weathers, P., Wiseman, P., Croen, P. and Sewell, D.H. (1990). *Hearing abilities of Down Syndrome*. Paper presented at the Biennial Conference on Human development, Richmond, VA. http://doi.org/10.1017/CBO9781107415324.004

Mason-Apps, E., Stojanovik, V., & Houston-Price, C. (2011). Early word segmentation in typically developing infants and infants with Down syndrome: a preliminary study. In *Proceedings of the 17th International Congress of Phonetic Sciences* (pp. 1334-1337).

Matsubara, M., Terasawa, H., & Hiraga, R. (2014). The effect of musical experience on rhythm perception for hearing-impaired undergraduates. In *Systems, Man and Cybernetics* (SMC), 2014 IEEE International Conference on (pp. 1666-1669). IEEE.

Næss, K. A. B., Lyster, S. A. H., Hulme, C., & Melby-Lervåg, M. (2011). Language and verbal short-term memory skills in children with Down syndrome: A meta-analytic review. *Research in Developmental Disabilities*, 32(6), 2225-2234.

Parbery-Clark, A., Anderson, S., Hittner, E., & Kraus, N. (2012). Musical experience strengthens the neural representation of sounds important for communication in middle-aged adults. *Frontiers in Aging Neuroscience*, 4, 30.

Parbery-Clark, A., Skoe, E., Lam, C., & Kraus, N. (2009). Musician enhancement for speech-in-noise. *Ear and Hearing*, *30*(6), 653–61. http://doi.org/10.1097/AUD.0b013e3181b412e9

Patel, A. D. (2008). *Music, language, and the brain*. Oxford university Press: Oxford.

Pekkonen, E., Osipova, D., Sauna-Aho, O., & Arvio, M. (2007). Delayed auditory processing underlying stimulus detection in Down syndrome. *Neuroimage*, 35(4), 1547-1550.

Peter, B., & Stoel-Gammon, C. (2008). Central timing deficits in subtypes of primary speech disorders. *Clinical Linguistics & Phonetics*, *22*(3), 171–98. http://doi.org/10.1080/02699200701799825

Peters, J. S. (2000). *Music therapy: An introduction*. (2nd Edition) Springfield, IL: Charles C Thomas, Publisher.

Pettinato, M., & Verhoeven, J. (2009). Production and perception of word stress in children and adolescents with Down syndrome. *Down Syndrome Research and Practice*, 1–13. http://doi.org/10.3104/reports.2036

Phillips-Silver, J. (2009). On the Meaning of Movement in Music, Development and the Brain. *Contemporary Music Review*, 28(3), 293–314. http://doi.org/10.1080/07494460903404394

Philips-Silver, J. and L.J. Trainor (2007), Hearing What the Body Feels: Auditory Encoding of Rhythmic Movement, *Cognition*, 105 (3): 533–46.

Phillips-Silver, J., & Trainor, L. J. (2005). Feeling the beat: movement influences infant rhythm perception. *Science*, 308(5727), 1430-1430.

Phillips-Silver, J., & Trainor, L.J. (2008). Vestibular influence on auditory metrical interpretation. *Brain and Cognition*, Vol. 67, pp. 94-102

Picard, B. M. (2009). Music and down's syndrome. Master's thesis, Royal Welsh College of Music and Drama. Retrieved from http://www.melodymusic.org.uk/publications/4574047824

Porter, H. L., Grantham, D. W., Ashmead, D. H., & Tharpe, A. M. (2014). Binaural Masking Release in Children With Down Syndrome. *Ear and hearing*, *35*(4), e134-e142.

Purser, H. R., & Jarrold, C. (2005). Impaired verbal short-term memory in Down syndrome reflects a capacity limitation rather than atypically rapid forgetting. *Journal of Experimental Child Psychology*, *91*(1), 1–23. http://doi.org/10.1016/j.jecp.2005.01.002

Rajendran, V., Roy, F. G., & Jeevanantham, D. (2012). Postural control, motor skills, and health-related quality of life in children with hearing impairment: a systematic review. *European Archives of Oto-Rhino-Laryngology*, 269(4), 1063-1071.

Reifinger Jr, J. L. (2006). Skill development in rhythm perception and performance: A review of literature. UPDATE: *Applications of Research in Music Education*, 25(1), 15-28.

Repp, B. H. (2005). Sensorimotor synchronization: A review of the tapping literature. *Psychonomic Bulletin & Review*, 12(6), 969-992.

Repp, B. H., & Doggett, R. (2007). Tapping to a very slow beat: a comparison of musicians and nonmusicians. *Music Perception: An Interdisciplinary Journa*l, 24(4), 367-376.

Repp, B. H., & Su, Y.-H. (2013). Sensorimotor synchronization: a review of recent research (2006-2012). *Psychonomic Bulletin & Review*, 20(3), 403–52. http://doi.org/10.3758/s13423-012-0371-2

Rigoldi, C., Galli, M., Mainardi, L., Crivellini, M., & Albertini, G. (2011). Postural control in children, teenagers and adults with Down syndrome. *Research in Developmental Disabilities*, *32*(1), 170–175. http://doi.org/10.1016/j.ridd.2010.09.007

Ringenbach, S. D., Allen, H., Chung, S., & Jung, M. L. (2006). Specific instructions are important for continuous bimanual drumming in adults with Down syndrome. *Downs Syndrome Research and Practice*, 11(1), 29–36. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/17048807

Rochette, F., Moussard, A., & Bigand, E. (2014). Music lessons improve auditory perceptual and cognitive performance in deaf children. *Frontiers in Human Neuroscience*, 8.

Roizen, N. J., Wolters, C., Nicol, T., & Blondis, T. A. (1993). Hearing loss in children with Down syndrome. *The Journal of pediatrics*, *123*(1), S9-S12.

Sacks, B., & Buckley, S. (2003). Motor development for individuals with Down syndrome–An overview. *Down Syndrome Issues and Information*. Retrieved from https://www.down-syndrome.org/information/motor/overview/

Sadakata, M., Hoppe, D., Brandmeyer, A., Timmers, R., Desain, P. (2008). Real-time visual feedback for learning to perform short rhythms with expressive variations in timing and loudness. *J. New Music Res.* **37**(3), 207–220

Saito, S. (2001). The phonological loop and memory for rhythms: An individual differences approach. *Memory* (Hove, England), 9(4), 313–322. http://doi.org/10.1080/09658210143000164

Schellenberg, G.E & Weiss, M.W. (2013). Music and cognitive abilities. In D. Deutsch (Ed.), Psychology of music (3rd Ed., pp 499-534). Elsevier

Schön, D., & Tillmann, B. (2015). Short- and long- term rhythmic interventions: perspectives for language rehabilitation. *Annals of the New York Academy of Sciences*, *1337*(1), 32-39.

Seung, H., & Chapman, R. (2000). Digit span in individuals with down syndrome and in typically developing children: temporal aspects. *Journal of Speech, Language, and Hearing Research*, 43, 609–620.

Shumway-Cook, A., & Woollacott, M. H. (1985). Dynamics of postural control in the child with Down syndrome. *Physical Therapy*, *65*, 1315–1322.

Silverman, W. (2007). Down Syndrome: cognitive phenotype. *Mental Retardation and Developmental Disabilities Research Reviews*, *13*, 228–236. http://doi.org/10.1002/mrdd

Sourtiji, H., Hosseini, S. M. S., Soleimani, F., & Hosseini, S. A. (2010). Relationship between motor and mental age in children with Down syndrome. *Iranian Rehabilitation Journal*, *8*(1), 4-7

Stephenson, J. (2006). Music therapy and the education of students with severe disabilities. *Education and training in developmental disabilities*, 290-299.

Stratford, B., & Ching, E. Y. Y. (1983). Rhythm and time in the perception of Down's syndrome children. *Journal of Intellectual Disability Research*, 27(1), 23-38.

Stratford, B., & Ching, E. Y. Y. (1989). Responses to music and movement in the development of children with Down's syndrome. *Journal of Intellectual Disability Research*, 33(1), 13-24.

Thaut, M. H. (2008). *Rhythm, music, and the brain: Scientific foundations and clinical applications*. Routledge.

Tierney, A., & Kraus, N. (2013). The ability to move to a beat is linked to the consistency of neural responses to sound. *The Journal of Neuroscience*, 33(38), 14981-14988.

Vicari, S. (2006). Motor development and neuropsychological patterns in persons with Down syndrome. *Behavior Genetics*, *36*(3), 355–364. http://doi.org/10.1007/s10519-006-9057-8

Vatti, Santurette, Pontoppidan, & Dau, T. (2014). Perception of a sung vowel as a function of Frequency-Modulation rate and excursion in listeners with normal hearing and hearing impairment. *Journal of Speech, Language, and Hearing Research*, 57(October 2014), 1679–1691. http://doi.org/10.1044/2014

Wang, J. H., Wang, S. A., Chen, W. C., Chang, K. N., & Chen, H. Y. (2012). Real-time pitch training system for violin learners. In *Multimedia and Expo Workshops (ICMEW), 2012 IEEE International Conference on* (pp. 163-168). IEEE

Welch, G. F. (1998). Early childhood musical development. *Research Studies in Music Education*, *11*(1), 27-41.

Welch, G. F., Howard, D. M., & Rush, C. (1989). Real-time visual feedback in the development of vocal pitch accuracy in singing. *Psychology of Music*, *17*(2), 146-157.

Welch, G.F., Howard, D.M., Himonides, E., Brereton, J. (2005). Real-time feedback in the singing studio: an innovatory action-research project using new voice technology. *Music Educ. Res.* **7**(2), 225–249.

Welch, G. F., Saunders, J., Edwards, S., Palmer, Z., Himonides, E., Knight, J. et al. (2015). Using singing to nurture children's hearing? A pilot study. *Cochlear Implants International*, 16(sup3), S63-S70.

Wilson, P. H., Lee, K., Callaghan, J. and Thorpe, C.W. (2008). Learning to sing in tune: Does real-time visual feedback help?. *Journal of interdisciplinary music studies* 2 (2008).

Wood, S. (2010). Electropalatography in the assessment and treatment of speech difficulties in children with Down syndrome. *Down Syndrome Research and Practice*, *12*(2), 98-102.

Yin, R. K. (2011). *Applications of case study research*. Sage.

Table 1: Cognitive measures
The table shows the cognitive measures that the participant undertook, the tests used, the scoring system, and the results

Cognitive measure	Test	Measure	Result
Verbal Mental-Age equivalent	British Picture Vocabulary Scale (BPVS) 1	Raw score	25
	BPVS 1	Age equivalent (years; months) and range (years; months)	3;0 m (2;8 - 3;5)
Non-Verbal Mental-Age equivalent	Draw-A-Person (DAP)	Age equivalent (years; months)	4; 3
Auditory memory	Digit Span	Max. number of digits correctly imitated	2
Phonological memory	Children's Test of Nonword Repetition (CToNWR)	Number of nonwords correctly imitated	0/10

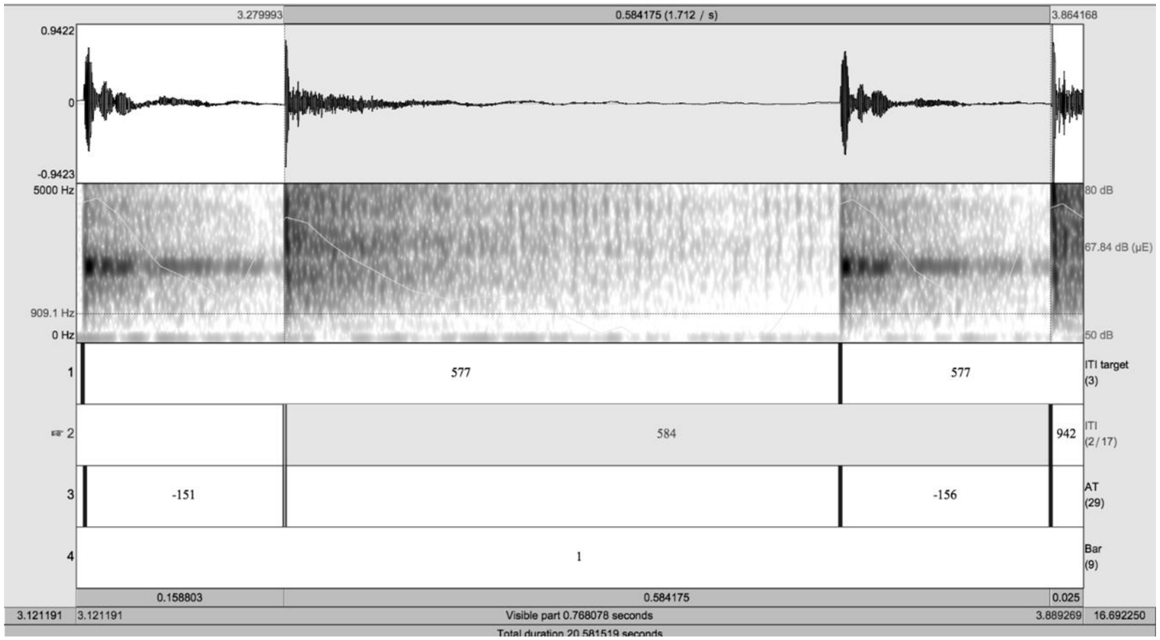

Figure 1: Calculation of the Beat Entrainment data using Praat. The text grid shows the waveform (top) and spectrogram of an extract of the beat entrainment task at 104 bpm. The duration between successive taps (ITI) in the target beat is marked in Tier 1, in milliseconds. The ITI of Robert's taps is marked in Tier 2, and the Anticipation Time (AT, ms) in Tier 3. Robert's taps were produced after the target beat, resulting in a negative AT. The Bar number is shown in Tier 4.

Table 2. Gross motor movement scores in aural and audio-visual conditions.

The table shows the scores allocated to Robert when reproducing (aural condition) or imitating continuous (visual assistance) gross motor movements to the song *Beat It* by Michael Jackson. The criteria and range for scores are given at the foot of the table.

Target motor movement, in time to the pulse	Aural condition only	Aural with ongoing visual assistance
Tap dominant foot	2	2
Step on the spot, alternating feet	0	0
Walk around the room	1	2
Lift both arms out to the side, and drop	1	2
Lift alternate arms out to the side, and drop	0	1
Clap hands	2	2
Raise both shoulders towards ears	2	2
Raise alternate shoulders towards ears	2	2
Twist torso	2	3
Independently, create a movement	2	n/a
Score: total/maximum score (%)	14/50 = 28%	16/45 =35.55%

Scoring:

0	1	2	3	4	5
no participation/unable to perform task	no regularity: wide variation between successive beats	maintains a regular tempo, but at own rhythm, not imposed	movements are mostly regular and are sometimes in time eg. for short time or with visual assistance. But lag behind/in front	movements are regular and coincide with most strong beats	accurate: feet coincide with beat

Table 3: Beat entrainment

Mean results for tapping to a metronome at tempos of 104 bpm, 132 bpm, and 160 bpm. The table shows the duration of mean Inter-Tap-Interval (ITI) and Anticipation Time (AT) in milliseconds, as a percentage of the target interval, and the SD in milliseconds. The + symbol indicates that Robert's taps occurred after target beats.

Measure	104 bpm	132 bpm	160 bpm
Target Inter-Tap-Interval (ITI), ms	576	455	375
No. of taps produced	20	25	33
ITI, ms	799	583	390
ITI, accuracy %	72	78	96
SD of ITI, ms	145	36	57
Mean range of ITI, accuracy =16%			
AT, ms	+184	+108	+199
AT, accuracy %	32	24	53
SD of AT, ms	191	159	73
Mean AT, accuracy =36%			

Robert's beat, ms			**1**		**2**		**3**		**4**		**5**	**6**		**7**		**8**		**9**		**10**
Target beat, ms	**1**		**2**	**3**	**4**		**5**		**6**		**7**	**8**	**9**		**10**					
Time, ms	0	280	560	840	1120	1500	1780	2060	2340	2620	2900	3180	3460	3740	4020	4300	4580	4860	5140	5420
Bar	4										5									

Figure 2. A timeline of the final 2 bars of Rhythm 1: *Ta tee-tee Ta Ta*. The timeline shows the approximate alignment of Robert's final ten beats in the last two bars of Rhythm 1 relative to the onset of ten Target beats, and the approximate duration (ms) of Robert's beats 1-9. The timeline is divided into equally-spaced units of 280 ms (line 3), which is the average duration of the quarter note (tee). Beat numbers are placed at approximate positions relative to sub-divisions of 140 ms. Robert's beats are longer in duration than target beats, and lag behind the stimulus. Robert was asked to stop playing at his Beat 10 and to 'watch my fingers'. Shaded areas exclude previous beats played by Robert, and continuing target beats for ease of comparison to the Target and to Figure 4.

Robert's beat, ms		**1**	**2**		**3**		**4**			**5**			**6**		**7**	**8**			**9**		**10**
Target beat, ms	**1**	**2**	**3**	**4**	**5**		**6**		**7**	**8**	**9**	**10**									
Time, ms	0	280	560	840	1120	1500	1780	2060	2340	2620	2900	3180	3460	3740	4020	4300	4580	4860	5140	5420	5520
Bar	3								4								5				

Figure 3. A timeline of bars 3-5 of Rhythm 2: *tee-tee tee-tee Ta Ta*. The timeline shows the approximate alignment of Robert's final ten beats in bars 3-5 of Rhythm 2 relative to the onset of ten Target beats, and the approximate duration (ms) of Robert's beats 1-9. The timeline (line 3) is divided into 280 ms, which is the average duration of the quarter note (tee). Beat numbers are placed at approximate positions, relative to sub-divisions of 140 ms. Robert's beats lag behind the stimulus. Although the duration of his beats is greater than the duration of target beats, the duration between beats 1-2 and 7-8 relative to his longer beats reflects the target 'tee-

tee' pattern. Shaded areas exclude previous beats played by Robert played in Bar 3 and exclude continuing target beats for ease of comparison to the target and to Figure 5. The task was concluded after the fifth target bar.

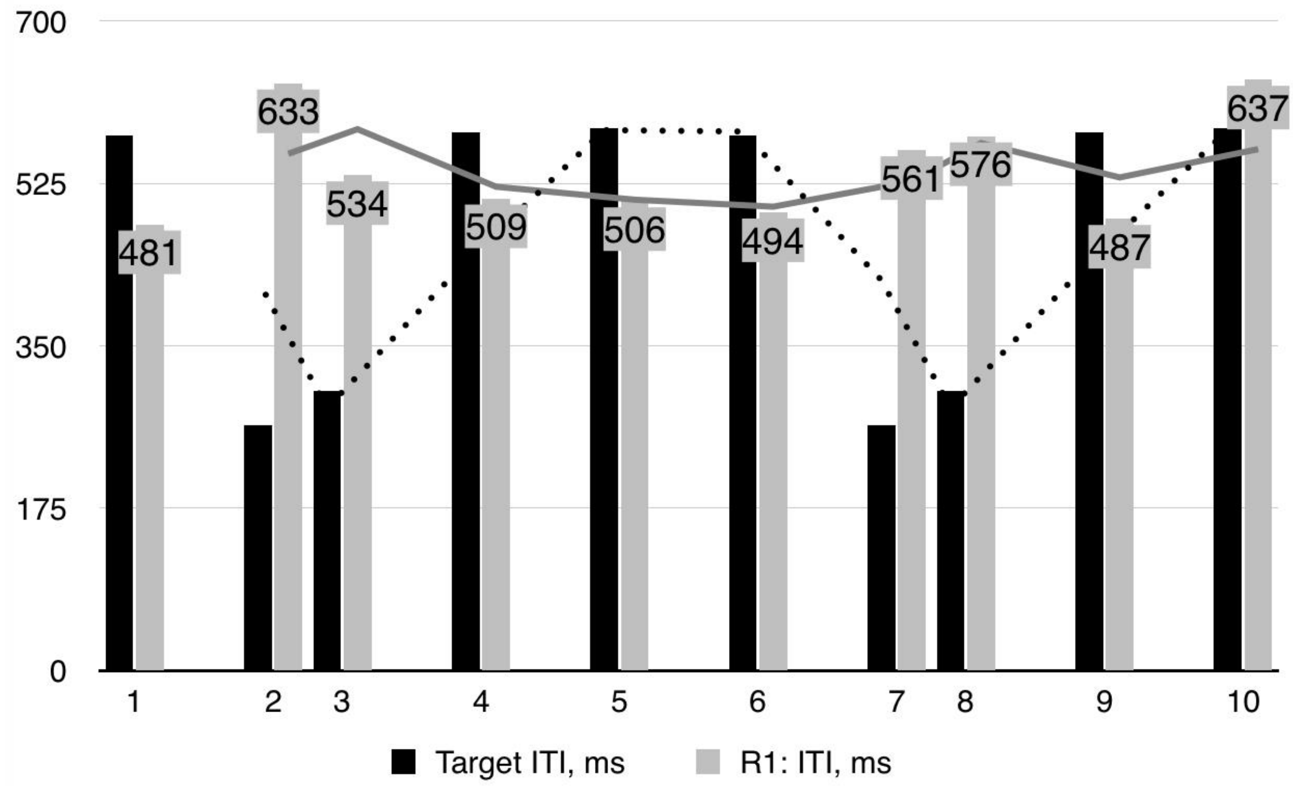

Figure 4: Graph showing the target ITI and the ITI of Robert's production in the Rhythm entrainment task - ta tee-tee ta ta. The chart shows the duration in milliseconds (ITI, ms: vertical axis) between beats (horizontal axis) of the task shown in Figure 2. The black bars show the target ITI (ms), which consists of a pattern of ten beats, in a 2:1 ratio. The grey bars represent the duration between Robert's beats, which were produced in response to the auditory stimulus only. The duration between his beats (ms) is marked at the

top of each column. The solid grey and dotted lines show the 'moving average' of Robert's production and of the target rhythm, respectively.

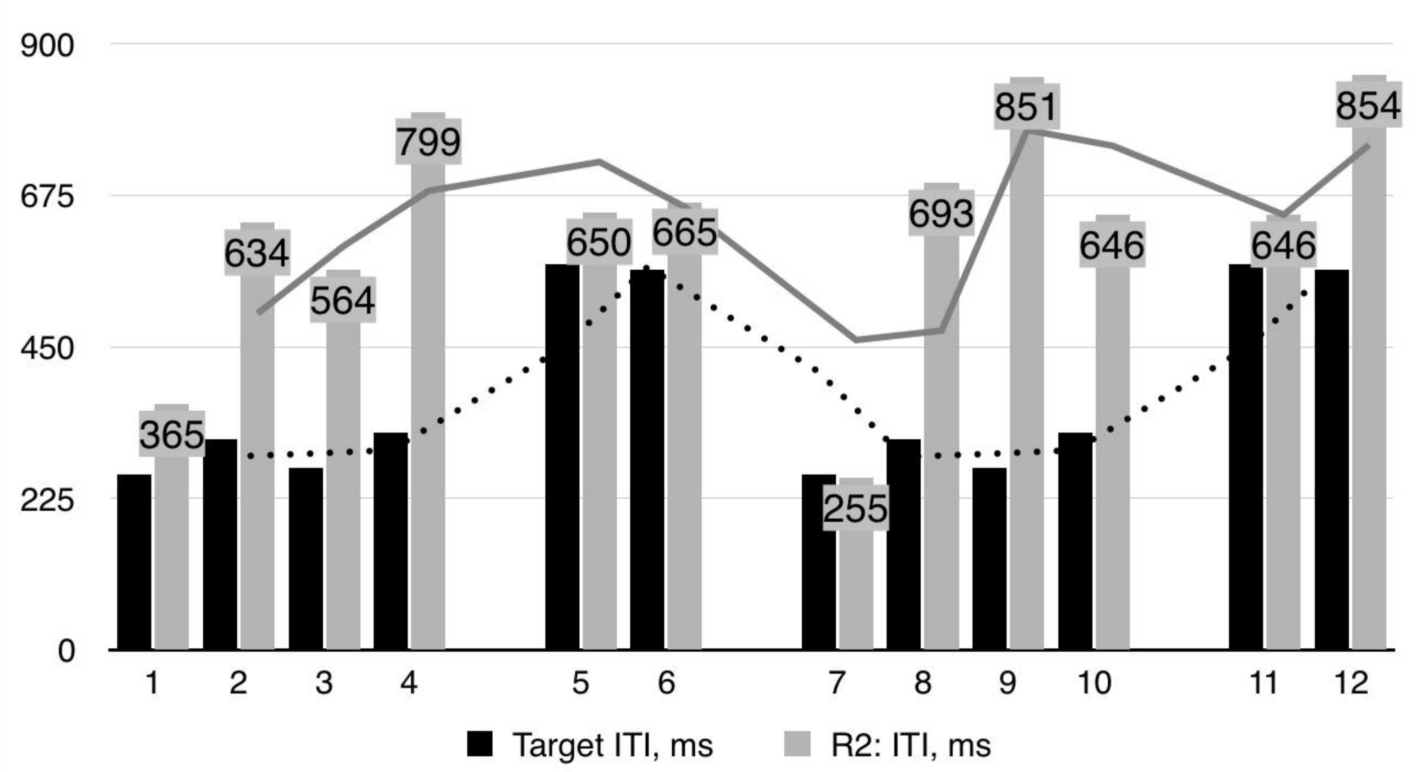

Figure 5: Graph showing the target ITI and the ITI of Robert's production in the second Rhythm entrainment task - tee-tee tee-tee ta ta. The chart shows the duration in milliseconds (ITI, ms: vertical axis) between beats (horizontal axis) of Rhythm 2 (see also Figure 3). The black bars show the target ITI (ms), which consists of a pattern of ten beats, in a 2:1 ratio. The grey bars represent the duration between Robert's beats, which were produced with ongoing visuo-spatial support. The duration between his beats (ms) is marked at

the top of each column. The solid grey and dotted lines show the ‘moving average’ of Robert’s production and of the target rhythm, respectively.