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Investigation of the neurological effects of musical training in children with Down Syndrome

Thesis Presented by
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For the degree of
Master of Science

University College Cork
Department of Anatomy and Neuroscience
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Supervisors: Prof. Yvonne Nolan, Dr. Jason Chan, Dr. Annalisa Setti

2024

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Declaration

All the work was my own unless stated. This research project was carried out under the supervision of Prof. Yvonne Nolan from the Department of Anatomy and Neuroscience, Dr. Jason Chan and Dr. Annalisa Setti of the School of Applied Psychology between January 2019 and March 2024. This thesis has not been submitted or partially submitted for any other degree or qualification.

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Eva McMullan, PhD.

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Author Contributions

The author carried out all research independently, except for the following contributions:

Dr. Jason Chan created the cognitive test programmes, including the Temporal Order Judgement task and the Corsi Blocks test.

Anna Gobleva and Gerard Moloney, both of whom helped process the saliva samples in the lab.

Padraig Wallace collaborated in creating the music intervention and its recording.

Don Slyne edited the videos for the intervention.

Maeve Hurley advised on creating the graphs.

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Abstract

The World Health Organisation (WHO) estimates that one in every 1000/1100 babies is born with Down Syndrome worldwide, with Ireland having the highest Down Syndrome occurrence than any other country, with 1-in-444 children being affected. It is characterised by physical growth delays, impaired sociability, anxiety symptoms, and intellectual disability. This cohort has differing capacities in executive function, attention, and learning and memory due to altered hippocampus and prefrontal cortex functions. Recent reports have suggested that music training could positively affect the cognitive and social abilities of children with Down Syndrome. However, we do not know if musical training improves sensory processing or higher-order areas in this cohort. This lack of understanding is detrimental to identifying the most effective means to enhance cognitive performance, relieve stress and improve the quality of life for those with an intellectual disability. Therefore, this study used a pre-post design to assess five types of cognitive processes, including learning and memory, sensory-motor skills, emotion regulation, social bonding, and executive function, before and after seven weeks of musical training. Saliva and hair samples were collected before and after the training sessions to examine concentrations of cortisol, which is linked to stress, and oxytocin, which is linked to social bonding. Temporal order judgement tasks, including both audio-visual blocks, were administered before and after the intervention period, as well as the Corsi Blocks test. In addition, all participants answered two questionnaires: Why Choose Music? (Paterson, 2008) and the Paediatric Quality of Life Inventory (Varni et al., 2001). The group that received the music intervention answered Daily Diaries and the Qualitative Description of Your Child's Experience of the Music Intervention questionnaire. Due to the challenges experienced with recruiting, the saliva and hair samples could not be processed. The results from the Temporal Order Judgement and the Corsi Blocks tasks did not show any significant difference in this cohort's temporal perception and short-term spatial memory. There was no improvement in short-term spatial memory and temporal perception as a result of the music intervention, suggesting that those cognitive domains

are not sensitive to a music intervention, at least as measured using these tasks. However, this could be because few participants completed the post-intervention measure. The other measures showed positive indicators of the emotional and social functions, as well as communication skills of those with Down Syndrome engaging with music. The diaries and the quantitative data saw sequential songs as the preferred musical activity when accompanied by actions. This led to perceived improvements in learning and memory that impacted communication skills in those with Down Syndrome.

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Abbreviations

ACh	Acetylcholine
ACORNS	Accessible Cause-Outcome Representation Notation System
ACTH	Adrenocorticotropic Hormone
ACTB	Arizona Cognitive Test Battery
AD	Alzheimer's Disease
ANOVA	Analysis of Variance
A-P	Anterior Posterior
ADHD	Attention Deficit Hyperactivity Disorder
ASD	Autism Spectrum Disorder
AVR	Aortic Valve Replacement
BADYG-E3	Battery of Differentials and Generals Aptitudes
BL	Blended Learning
BLT	Bright Light Therapy
bpm	Beats Per Minute
BRIEF-P	Behaviour Rating Inventory of Executive Function-Preschool
CA	Chronological Age
CABG	Coronary Artery Bypass Grafting
CANTAB	Cambridge Neuropsychological Test Battery
CDI	Communicative Development Inventory
CES	Central Executive System
CFC	Contextual Fear Conditioning
CiPS	Composers in Public Schools
CPC	Cortico-Ponto-Cerebellar
CREC	Clinical Research Ethics Committee
CRH	Corticotropin-Releasing Hormone
CS	Conditional Stimulus
CTC	Cerebello-Thalamo-Cortical (Yvonne marked this)

DA	Developmental Age
DG	Dentate Gyrus
DH	Dorsal Hippocampus
dLPFC	Dorsolateral Prefrontal Cortex
dMPFC	Dorsomedial Prefrontal Cortex
DS	Down Syndrome
DSE	Down Syndrome Education
DTBM	DTI-driven tensor-based morphometry
EC	Entorhinal Cortex
ECG	Electrocardiogram
EF	Executive Function
EEG	Electroencephalogram
EFL	End Folium
Ep	Ependymal surface
FMRI	Functional Magnetic Resonance Imaging
fNIs	Functional Neon-Infrared Spectroscopy
ff	Fortissimo
GCL	Granular cell layer
HATA	Hippocampal-Amygdaloid Area
HCG	Hair Cortisol Concentration
HG	Heschel's Gyrus
HM	Henry Molaison
HPA	Hypothalamus-Pituitary Adrenals
HPC	Hippocampus
HR	Heart Rate
HRQOL	Health-Related Quality of Life
HRV	Heart Rate Variability
Hsa21	Human Chromosome 21
IBRS-R	Infant Behaviour Rating Scale-Revised

ID	Intellectual Disabilities
IH	Inferior Horn
IQ	Intelligence Quotient
IV	Intravenous
LEEIP	Little Explorers Early Intervention Programme
LI	Language Impairment
LTM	Long Term Memory
LTP	Long Term Potentiation
MA	Mentally Aged
MAGL	Music Appreciation Guided Listening Curriculum
MCP	Middle Cerebellar Peduncle
MINdDS	Music as an Intervention in the development of children with Down Syndrome
MoCA	Montreal Cognitive Assessment
MRI	Magnetic Resonance Imaging
MSB	Music and Social Bonding Hypothesis
MTL	Medial Temporal Lobe
NAcc	Nucleus Accumbens
NEPSY	A Developmental Neuropsychological Assessment
OECD	Organisation for Economic Co Operations and Development
oFPFC	Orbitofrontal Prefrontal Cortex
OXTR	Oxytocin Receptors
PANAS	Positive and Negative Affect Schedule
PaO2	Partial Pressure of Oxygen
PaS	Parasubiculum
PCL	Pyramidal Cell Layer
PedsQL	The Paediatric Quality of Life Inventory
PFC	Prefrontal Cortex
pp	Pianissimo
PPVT	Peabody Picture Vocabulary Test

PRL	Prolactin
PrS	Presubiculum
PSS	Perceived Stress Scale
PVN	Paraventricular Nuclei
R	Rest without Stimulation
RAM	Radial Arm Maze
RM	Relaxing Music
RSA	Respiratory Sinus Arrhythmia
sAA	Salivary alpha-amylase
SaO2	Arterial Oxygen Saturation
SCP	Superior Cerebellar Peduncle
SIC	Symptoms of Illness Checklist
SIFI	Sound-Induced Flash Illusion
SLT	Speech and Language Therapy
SOA	Stimulus-Onset Asynchrony
SON	Supraoptic Nuclei
SSM	Spatial Sequential Memory
SoHT	Society of Hair Testing
STG	Superior Temporal Gyrus
STM	Short Term Memory
STSA	Storytelling and Story-acting
STVSM	Short Term Visuospatial Memory
SW	Sound of Rippling Water
SWM	Spatial Working Memory
TD	Typically Developing
TMS	Transcranial Magnetic Stimulation
TOJ	Temporal Order Judgement
TSST	Trier Social Stress Test
US	Unconditional Stimulus

VH	Ventral Hippocampus
vLPFC	Ventrolateral Prefrontal Cortex
vMPFC	Ventromedial Prefrontal Cortex
ViSM	Visual Spatial Memory
VSTM	Visual Short-Term Memory
VSWM	Visuospatial Working Memory
VTA	Ventral Tegmental Area
VWM	Visual Working Memory
WHO	World Health Organisation
WISC-IV	Wechsler Intelligence Scale for Children, Fourth edition
WM	Working Memory

General Introduction

1.1 What is Down Syndrome?

Down Syndrome (DS) is a common genetic condition that affects 8% of all registered births in Europe, with 1 in approximately every 444 babies affected by the syndrome in Ireland, resulting in 110 babies being born with DS annually (Windsperger & Hoehl, 2021). The World Health Organisation (WHO) estimates that one in every 1000/1100 babies is born with DS worldwide, with Ireland having the highest DS occurrence of any other country (Rodrigues, 2019). The global population of people with DS is estimated at 6 million (Hanney, Prasher et al., 2012).

Down Syndrome involves the triplication of the Human Chromosome 21 (Hsa21), which results in the overexpression of more than 500 genes. While detected initially by Charles Darwin in the *Origin of the Species*, it was named after John Langdon Down in 1866. Langdon Down was the first to conduct pioneering research that differentiated this cohort from other groups with intellectual disabilities (ID) (Lott & Dierssen, 2010). However, it was not until 1959 that Jérôme Lejeune discovered that DS was a consequence of the trisomy of chromosome 21 (Neri, 2009).

A physical map of chromosome 21 was completed by Hattori et al. in 2000. Here, the researcher discovered 225 gene variants that contribute to the neurological and physical complexities of DS (Hattori, Fujiyama et al., 2000). To date, several large-scale studies have investigated the many health complications associated with DS, making it a multi-disciplinary field of study (Turner, Sloper et al., 1990; Yam, Tse et al., 2008; Schieve, Boulet et al., 2009; Schieve, Boulet et al., 2011). The clinical features include a characteristic facial profile, a short stature, a flatness in the nasal cavity, and an extended tongue (Windsperger & Hoehl, 2021). People with DS are at a higher risk of being affected by celiac disease, diabetes, respiratory issues, hypothyroidism, and cardiovascular disease, while half of the infants born with DS have congenital heart conditions (Freeman, Taft et al., 1998; Roizen & Patterson, 2003). Additionally, many of those with DS have the potential to experience hearing and visual impairments (Turner, Sloper et al., 1990; Marcell & Cohen, 1992). McCaughey et al. have found that sleep apnoea and obstructive sleep conditions also affect this cohort (Levanon, Tarasiuk et al., 1999; Carter, McCaughey et al., 2009).

As a result of advanced medical treatments and interventions now available, the life expectancy of people with DS has increased dramatically, from 12 years of age in the 1940s to 60 years (Zigmond, Rowland et al., 2015). De Graaf et al. cite the most recent figures compiled in 2010 showing that 57% of people with DS at that time were older than 20 years, while 28% of the population were older than 40 years (Graaf et al., 2021). O'Shea and Pierce predict that the next generation of people with DS will have the same life expectancy as typically developing (TD) adults (Cahill, O'Shea et al., 2012). With an optimistic lifespan for the DS community, understanding the functional strengths and abilities of this cohort is imperative in creating suitable learning opportunities and supports that can improve functionality and, hence, their quality of life. While DS is one of the 'oldest' syndromes to be discovered, very little is known about its neuropsychology (Pennington, Moon et al., 2003).

Varying levels of cognitive ability are experienced by people with DS. Cognition refers to various higher mental processes that enable people to function efficiently in society. These include thinking, perceiving, imagining, speaking, acting, and planning (Ward, 2015). The many altered cognitive abilities result from differences in the brain formation of people with DS, including structural, volumetric, and connectivity between different brain regions (McCann, Levman et al., 2021). These neurological differences have, in turn, impacted the roles of primary neurocognitive domains, including language, attention, memory, executive function (EF), and social cognition. These affected areas can impact higher-order mental processes involved in planning and decision-making that can significantly influence the ability of people with DS to interact in social situations (Harvey & Penn, 2010; Amadó, Serrat et al., 2016).

The proposed research project, Music as an Intervention in the Neurologic development of children with Down Syndrome (MINdDS) stems from musical training already in use with a group of children in Cork that was created by the author, which has self-reported effectiveness by parents of enrolled children with DS. Recent data has suggested that music training can positively affect children of all abilities; however, it remains unclear if it improves sensory processing or higher-order areas (Miendlarzewska & Trost, 2014). This lack of understanding is detrimental to identifying the most effective means to enhance cognitive performance, relieve

stress, and improve the quality of life for those with ID. Musical training can potentially improve behavioural outcomes in children with cognitive disorders, but the specific benefits obtained are not well understood. However, there is value and potential in knowing more about music training techniques and how they impact at a neurological level. Therein lies the motivation for the following thesis: to further examine the abilities of this population in response to an appropriate music programme that could strengthen these existing capacities.

1.2 Mediating Medical and Social Models of Disability

Both medical and social models of disability are often put at either end of the spectrum when talking about disability. The medical model is based on the biomedical impairments responsible for a particular disability (Hargrave, 2017). Therefore, it has traditionally been associated with identifying the deficits of someone with a disability and how these aberrations might be rectified or fixed. The view has often been that the person with a disability is portrayed as being in some way 'lesser' through this model by highlighting the deficits and emphasising the negative impacts. In contrast, traditionally, the social model is person-centred and identifies how society is responsible for the barriers that have prevented people with disabilities from excelling. As specified in a seminal publication, *Theatres of Learning Disability: Good, Bad, or Plain Ugly?* by Hargrave, there are significant limitations to adopting a social model exclusively (Hargrave, 2017). The model embraced by this thesis aims to acknowledge the importance of considering aspects of both models to establish a learning profile of people with DS and, hence, enhance or inform the teaching methodology of an intervention (Pickard, 2019). Inspired by a recent article by Pickard, 'Mediating Medical and Social Models of Disability in Instrumental Teaching for Children with Down Syndrome' (Pickard, 2019), this thesis similarly acknowledges the importance of both models to understand the challenges, the reasons why these challenges exist, and how the strengths of this cohort can lead to more effective methods of teaching. The negativity implied by the medical model is outdated and acts only to acknowledge the physical limitations experienced by this cohort so that an informed response can be made. Celebrating ability is at the core of this research. It is the author's experience that ignoring the challenges only puts

additional barriers to finding appropriate methods of engaging children with DS. The current understanding of the developmental profile of these children will form the focus of the following sections.

1.3 Frameworks for Considering Atypical Developmental

How a child develops is multifaceted and trying to represent this cohesively and coherently within an efficient framework is often challenging in scientific studies. These challenges are compounded even further when describing the development of neurodiverse children. The available models tend to emphasise the deficiencies in these children rather than their abilities. Therefore, finding a suitable model that reflects the ethos of this project has been complex, as there are many contrasting models of approach to child development in psychology, neuroscience, and music. Several existing models address some aspects of development, but rarely in a comprehensive and multi-disciplinary approach that reflects the nature of this project (Inhelder & Piaget, 1958; Erickson, 1959; Bowlby, 1968; Bandura, 1980; Vygotsky, 1981). Therefore, aspects of Morton's causal developmental model (2004) and the neuroconstructivist model (Karmiloff-Smith, 1998). will be proposed as approaches that could help capture and explain cognitive and anatomical features in those with DS in the mediated territories between the medical and social models of disability.

1.3.1 A Structural Causal Model for Developmental Psychopathology

Nel mezzo del cammin di nostra vita mi ritrovai per una selva oscura ché la diritta via era smarrita. Dante

One of the models that has contributed to the theoretical framework applied to the current research project is Morton and Firth's causal model of development (Morton &Firth, 1995). Found at the start of an article by Morton and Frith in 1993, the opening citation resonates with the challenges of detailing the developmental trajectory of a child with DS, where "in the middle of the journey of my life, I found myself in a dark forest because the straight path was lost" (Morton & Frith, 1995). At the commencement of their article, Morton identifies that atypical development is often viewed as unilateral, disregarding other interrelated aspects of a condition. This

means that experts tend to focus on their respective fields rather than address the multifaceted nature of development. For example, medical researchers focus on the biological aspect, while psychologists focus on behaviour. Morton and Frith have proposed a third level that connects these two aspects, making it more applicable to their autism neurodevelopmental research (Frith, Morton et al., 1991). The diagrammatic nature of their approach shows distinctive connections being made effectively that linguistically would be more complex to depict.

Morton's causal model includes a tripartite structure when assessing complex development. This structure contains biology (brain construction), which influences cognition and the combined effect on behaviour. As seen in Figure 1.1, this causal model acknowledges the impact of environmental factors on this top-down approach (Moore & George, 2011). Building on the original causal model, the authors outlined the need to create a comprehensive visualisation system to represent the development of those with intellectual difficulties.

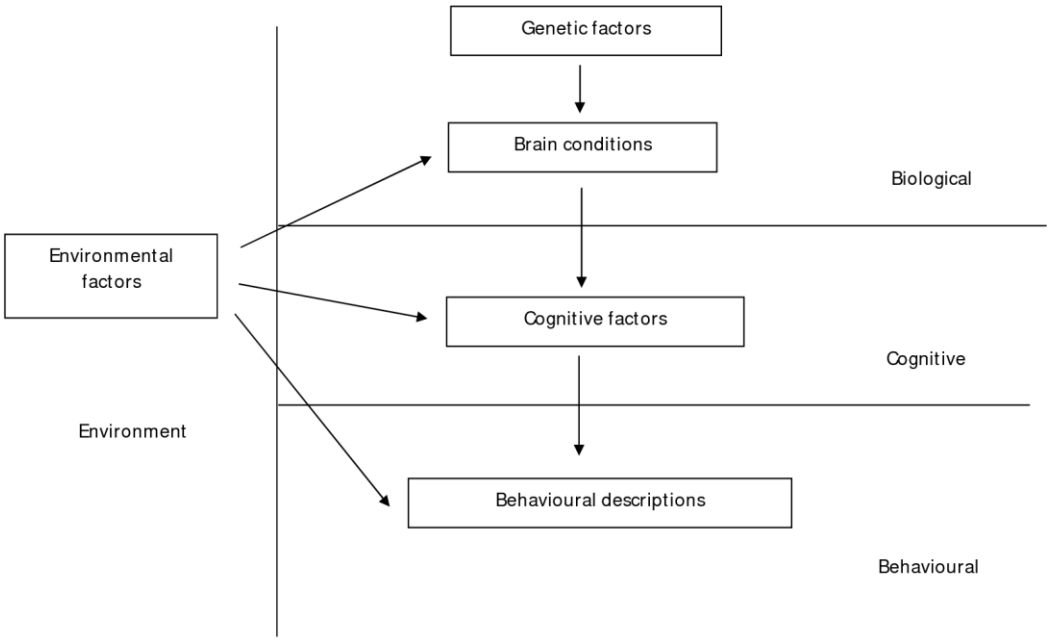


Figure 1.1. A tool for visualising and modelling atypical development (Moore & George, 2011), in D. G. Moore & R George (2011). ACORNS: a tool for the visualisation and modelling of atypical development. *J Intellect Disabil Res*, *55*(10), 956–972. https://doi.org/10.1111/j.1365-2788.2011.01471.x Reproduced with permission from *Wiley & Sons Books* (Blackwell Publishing Ltd.).

An Accessible Cause-Outcome Representation Notation System (ACORNS) was also designed so that clinicians and researchers working with someone with ID would have a standard procedure and language across levels of communication and, therefore, translatable across disciplines (Moore & George, 2011). The aim was to visually present atypical developmental profiles of those with ID. It is shown in a frame, progressing horizontally from left to right. The authors have expanded the original three categories of biology, cognition, and behaviour previously seen in Figure 1.1 to include the physical and social environment, offering a more comprehensive view of environmental conditions and their impact on development. The recognition of multiple impacts on development, including internal and external elements, makes this aspect of the causal model of development relevant to this current study of children with DS. The biological, cognitive, and behavioural elements are most often recognised when studying children with DS; however, the importance of how these aspects interact with the environmental conditions has yet to be considered in studies to date. How these elements act together forms a focus in the elemental design of the intervention.

However, while providing static representation, modular methods also tend to focus on recognising impairments more than strengths, which conflicts with the

ethos of this study. This causal model will be used primarily as an organisational strategy in conjunction with the following neuroconstructivist model staying within the mediated territory of medical and social models of disability.

1.3.2 Neuroconstructivist Model of Development

The neuroconstructivist model has also informed the theoretical framework for this current study, as it lends itself to analysing neurodevelopmental disorders more holistically. As proposed by Karmiloff-Smith, the growth process heightens our understanding of development (Karmiloff-Smith, 1998). In contrast to the previous model, neuroconstructivism takes a similar viewpoint as the Piagetian opinion that a child's development is an expansion of mental representations through experience-dependent processes (Westermann, Thomas et al., 2010). It is necessary to look through the 'lens' of how a child grows and develops within its environment (Gur, 2014). Unlike Piaget's theory of cognitive development, there are no development stages, but developmental changes in competencies are built with experience grounded in brain development and plasticity (Ward, 2015). However, in addition to Piagetian theory, neuroconstructivism is influenced by functional brain development, as it plays a pivotal role in cognition. This model acknowledges how the brain networks and connections change through action. The constant multilateral interaction between the genes, brain, body, and environment is detailed in Figure 1.2, which is central to the neuroconstructivist model and has become an integral framework for the current study.

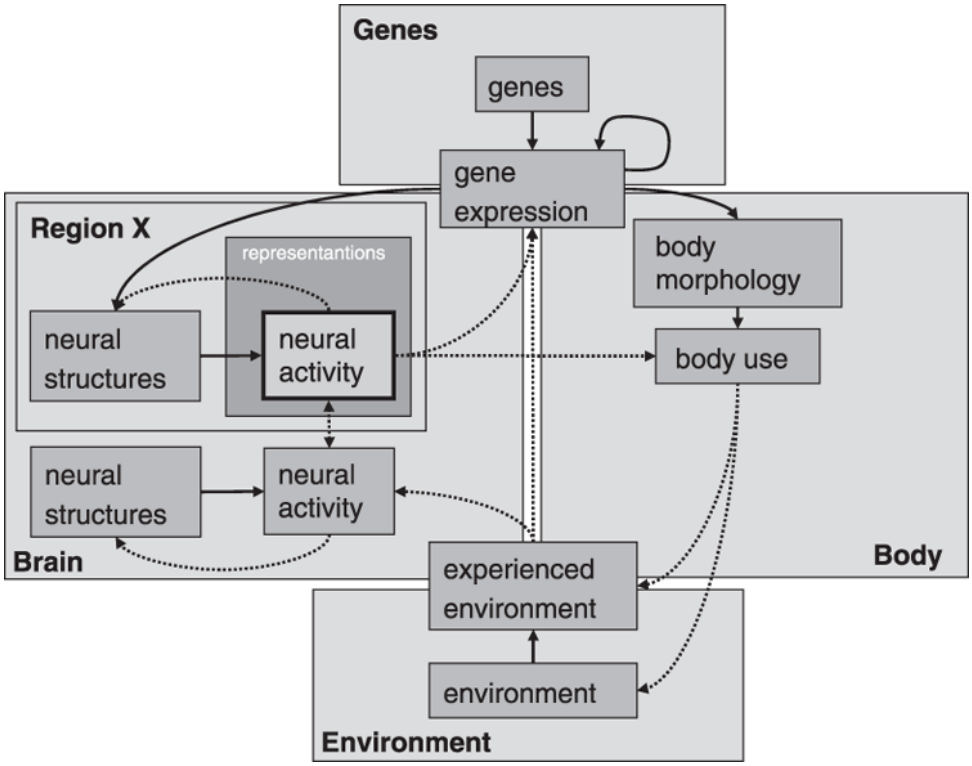


Figure 1.2. The neuroconstructivist model (Westermann, Mareschal et al. 2007). G. Westermann, G, D. Mareschal, M. Johnson, S Sirois, M. Spratling & M. Thomas. (2007). Neuroconstructivism. *Developmental Science*, *10*, 75–83. https://doi.org/10.1111/j.1467-7687.2007.00567.x Reproduced with permission from *John Wiley & Sons Books* (Blackwell Publishing Ltd.).

Westermann emphasises the importance of genes and bi-directional flow, which Gottlieb identifies as probabilistic epigenesis: the genes' intrinsic and extrinsic interaction with the environment (Gottlieb, 2007). There are common principles that this model adheres to, one of which is context-dependent; the biological conditions shape the developing neural system, which, in turn, creates a context that impacts development.

The neurocontructivist view is that functionally specialised brain regions are context-sensitive and depend on the interactions with other brain regions (Mechelli, Price et al., 2001). Therefore, the organisation of the cortical areas is due to the connections that have been formed. The neuroconstructivist view of development results from the interaction between genes with the environment and the mature cognitive system built from earlier interactions (Ward, 2015). This is understood through competition, which leads to specialisations in the system, cooperation that allows knowledge and information to be used at a more advanced level and chronology, which is associated with temporal constraints of development (Westermann, Thomas et al., 2010).

The neuroconstructivist approach provides a more malleable model that lends to discussing atypical development. In contrast to other models, it does not focus on deficits in a particular domain per this study (Farran, Courbois et al., 2012). In Farran's view, comparing parts of the brain that are 'intact' to defective parts gives little insight into the development trajectory of a child (Farran, Courbois et al., 2012). Farran believes that the brain develops sensitively through time, displaying subtle changes in cognition and behaviour that should be considered in context (Farran, Courbois et al., 2012). This coincides with Westermann, who believes that characteristics of atypical development are the same as typical development. Still, the conditions are different, leading to some abilities being heightened more than others (Westermann, Thomas et al., 2010). Therefore, development is context-dependent, and when atypicalities exist in the brain, they can impact other areas, leading to a high level of adaptation that allows people to interact with their environment in atypical but optimal ways (Franks and Richardson, 2006).

The acknowledged bidirectional interactions between the brain, genes, the body, and the environment are integral considerations in the approach taken in this current study. The importance of viewing atypical developments within a context without identifying deficits in a child's ability has also informed the theoretical framework devised for this research project. How the causal model of development and the neuroconstructivist approach have been aligned to create a development model for children with DS will form the focus of the following section.

1.3.3 Models of Development and Down Syndrome

Having delivered a music programme for Down Syndrome Cork (DSC) for several years, the researcher and the parents observed some interesting responses and developments in the children. The present study stemmed from these observations where it was felt that the abilities of children with DS were augmented through music engagement, therefore focusing on their strengths. While these were anecdotal outcomes, the parents felt a need to validate these findings scientifically. With a lack of significant research in the area (Gemma, Pablo et al., 2020), there was a need to emphasise how a carefully devised music programme could potentially strengthen specific abilities in this cohort. Such a project could offer insight into the

developmental trajectory of children with DS, provided a relevant model could be applied.

Cebula attempted to combine the neuroconstructivist and causal developmental models in an article addressing the social cognition in children with DS (Cebula, Moore et al., 2010). In looking at the social understanding of children with DS, much of the focus was on toddlers and infants who developed social skills on a par with their TD peers (Cicchetti, 1990). Cebula believes individual differences exist in children with DS and how they respond to certain social situations (Cebula, 2010). These subtle differences could be significant as they influence socio-cognitive and language development (Cebula, Moore et al., 2010). While many studies have attempted to depict the developmental trajectory of children with DS, as indicated by Cebula, the research still has considerable voids (Cebula, Moore et al., 2010). Cebula poses a theoretical framework that would lead to a better understanding of the socio-cognitive profiles of people with DS. As a result, an improved theoretical framework for understanding these aspects of the neurodevelopmental profile of this cohort could be better supported and understood. The researcher also acknowledges the inherent position of the parent and family network in aiding development, therefore supporting transactional models that place infants, caregivers, and their interaction with the environment as integral in behavioural outcomes (Cebula, Moore et al., 2010).

The model, however, shows a significant disconnect between the neurobiology of the children and other aspects of the model. While there is more multi-directional movement between social environment and social behaviour, there is still a top-down approach. Mapping the trajectory over time does not apply to the current study due to the duration limitation of the intervention; therefore, a modified association of these models is required. Thus, the causal model will function as an organisational strategy in what follows; however, the neuroconstructivist model will help support the analytical approach used in the intervention. The suggested neuro-causal model used within the intervention will be detailed in the following section.

1.3.4 Neuro-Causal Model for Atypical Development

While unable to assess how children with DS will develop over time within this study, understanding the constraints on neural development in children with DS and how it will help us understand cognition in this cohort is central to the project (Westermann, Mareschal et al., 2007). The importance of a positive collaboration with a child's surroundings and how this can influence development is also at the core of this research study. A child's inherent characteristics are because of an interaction between their genetic/neuronal makeup and their environment. Prioritising one over the other is counterproductive, but following research into epigenesis, the interaction between behaviours and the environment can cause changes that affect how our genes work.

Therefore, the original diagrammatic presentation of the previously proposed developmental framework by Westermann and Mareschal et al. (2007) has been adapted while also including aspects of the causal model used by Cebula and Moore (2011). The fundamental components of the Brain, Genes, Body, and Environment have been retained from the original model proposed by Westermann and Mareschal, while additional causal elements have been included. The role of physical and social environments in development and the significance of the temporal environment has been acknowledged. The temporal environment refers to the timing and sequence of events within a specific environmental context. The terms proactivity and progress have again been reused from the diagram by Westermann. The horizontal progression is in keeping with the ideology of the causal model introduced by Moore and Cebula. Finally, representation in cognition, both hot and cold, and behavioural have been included as outcomes. This approach accommodates cross-interaction between the contextual elements, with the assumed proactivity and progress being central regardless of the directionality of the model, as seen in Figure 1.3. This neuro-causal model also sits appropriately between the medical and social models of disability.

This neuro-causal model has formed the foundation of the intervention at all levels. The context of the participants, including their biological and environmental situations, directed the content of the intervention. At the same time, the

developmental processes were guided by the practice model used to deliver the programme. The outcomes were measured only under the acknowledged fluidity between each component.

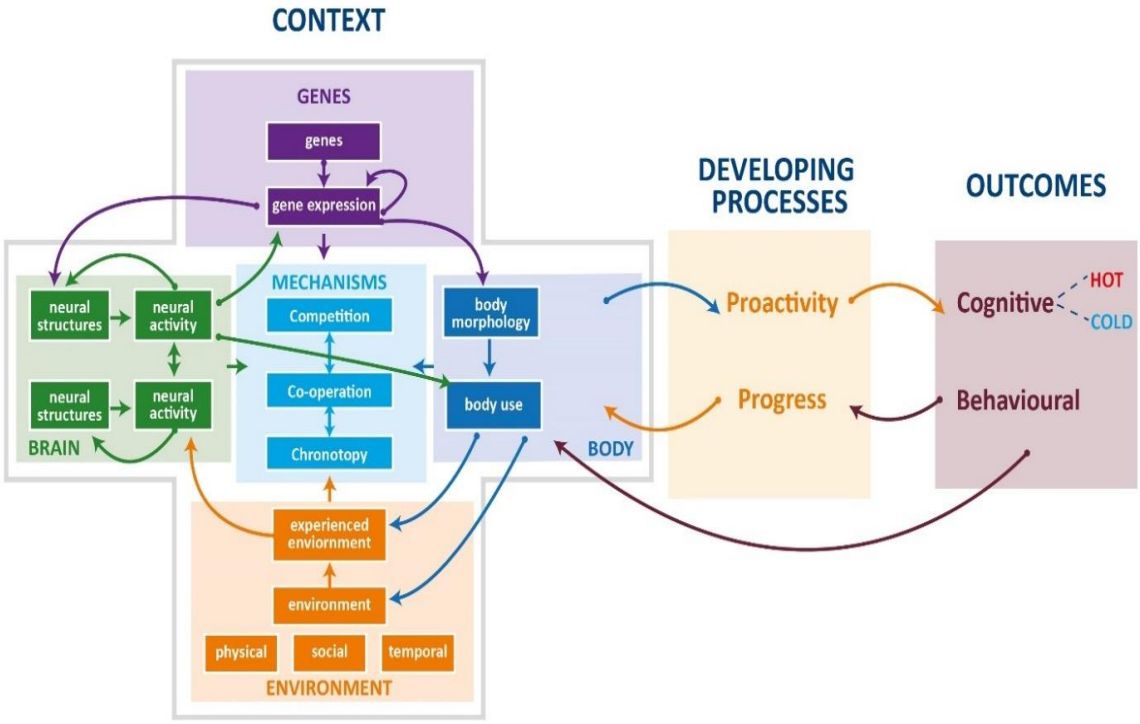


Figure 1.3. The neuro-causal model for considering atypical development is adapted from the neuroconstructive model (Westermann & Mareschal, 2010) and the causal model (Cebula, 2010; Moore & George, 2011).

1.4 Neuroanatomic Brain Structure: Children with Down Syndrome in Context

Down Syndrome manifests at the prenatal stage of development, and the foetus develops abnormally *in utero*. By 15 weeks of gestation, the brains of children with DS are significantly smaller than those of TD neonates at this embryonic age. While these developmental differences are evident at this stage, babies are rarely detected as having DS before birth without an amniocentesis. The brain of a DS baby weighs less than its TD counterparts, which has encouraged much research into the developmental trajectory of the brain in this cohort (Cowie, 1973). A reduced intracranial structure results in volumetric differences, mainly in the cerebellum, brainstem, and frontal lobes (Kesslak, Nagata et al., 1994; Raz, Torres et al., 1995; Aylward, Li et al., 1999). Studies have similarly shown decreased volume in the cingulate gyrus, medial temporal lobe, superior temporal lobes, and hippocampi

(Aylward, Li et al., 1999; Pinter, Brown et al., 2001; White, Alkire et al., 2003; Carducci, Onorati et al., 2013) with an independent study subsequently showing reduced grey matter in the inferior cerebellum, fusiform gyrus, and medial temporal lobe (Menghini, Costanzo et al., 2011). There is an evident increase in the size of the parahippocampal gyrus found in these babies compared to TD infants (Kesslak, Nagata et al., 1994).

The over-expression of chromosome trisomy 21 results in fewer neurons segregating to incorrect cortical layers (Kim, Kim et al., 2017). Neuronal apoptosis or 'cell death' occurs to a greater extent in both prenatal and postnatal babies with DS. Specifically, babies with DS show a 20%–50% reduction in neuronal density from 0–60 months, and this decrease in neurogenesis (neuronal birth) begins at 22 weeks of gestation (Schmidt-Sidor, Wisniewski et al., 1990). Consequently, delayed myelination (producing myelin sheaths around neuronal axons, which controls neuronal firing) results in neurotransmission differences. Studies have shown that infants' brains also show degeneration of the cortical pyramidal neurons, weakening dendrites and synapses in the hippocampus (HPC) and fewer granular cells in the cerebellar cortex (Zigmond, Rowland et al., 2015). Evidence shows a shortening in dendritic length and cerebral cortical pyramidal cells with fewer synapses, resulting in shorter local connections than inter-hemispherical connections and a lack of cross-hemispherical communication (Lott, 2012).

Consequently, the developing brain structures show disproportionate impairments, particularly in the cerebellum, which has 75% less volume than those found in the brains of TD children, and cerebellum development is delayed. Cerebellar alterations in children with DS may account for difficulties in fine motor skills and cause hypotonia. The cerebellum coordinates voluntary movement and balance and has been linked to EFs in recent literature (Beuriat, Cohen-Zimerman et al., 2020). A decreased brain volume and brachycephaly (shorter skull) (Figures 1.4 and 1.5) account for many intellectual challenges reported in children with DS.

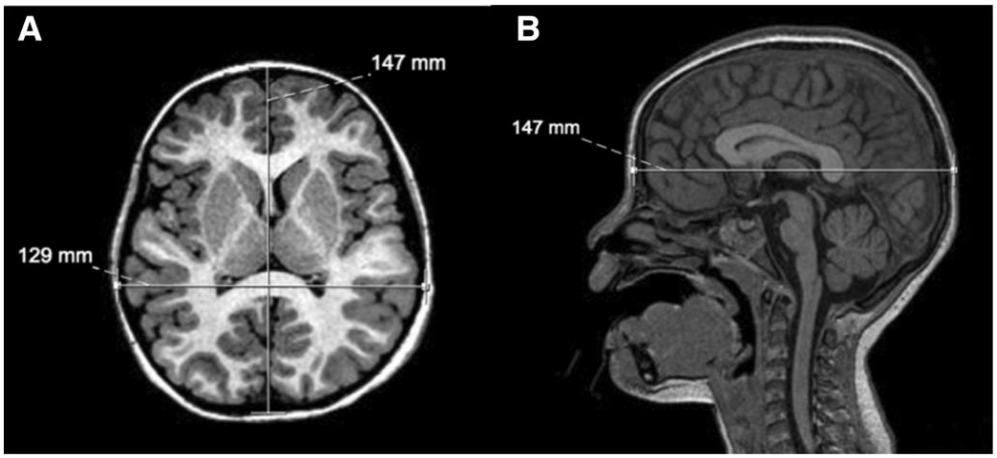


Figure 1.4. The neuroimaging of a 5-year-old child with Down Syndrome (Rodrigues, Nunes et al., 2019). M. Rodrigues, J. Nunes, S. Figueiredo, A. Martins de Campos, & A. F. Geraldo (2019). Neuroimaging assessment in Down syndrome: a pictorial review. *Insights into Imaging*, *10*(1), Article 52, 1–13, https://doi.org/10.1186/s13244-019-0729-3. Reproduced with permission from *Springer Nature*. (Creative Commons Attribution 3.0 License).



Figure 1.5. 3D reconstruction of a 5-year-old child with DS showing brachycephaly (Rodrigues, Nunes et al., 2019). M. Rodrigues, J. Nunes, S. Figueiredo, A. Martins de Campos, & A. F. Geraldo, A. F. (2019). Neuroimaging assessment in Down syndrome: a pictorial review. *Insights into Imaging*, *10*(1), Article 52, 1–13, https://doi.org/10.1186/s13244-019-0729-3. Reproduced with permission from *Springer Nature*. (Creative Commons Attribution 3.0 License).

A summary of the affected brain regions of those with DS is seen in Figure 1.6, with these areas influencing information acquisition, information storage, information processing, and information retrieval (Lott & Dierssen, 2010). The medial temporal lobe, specifically the HPC and the cerebellum, develops and matures postnatally. Therefore, these brain regions responsible for motor skills, memory, and learning are particularly at risk in this cohort (Nadel, 1986). People with DS account for 15% of the population with ID (Zigmond, Rowland et al., 2015). While some individuals with DS have an intelligence quotient (IQ) that falls within the normal range (Pueschel, 1995; Chapman & Hesketh, 2000), it is believed that approximately 80% of this population

have a moderate ID while others have a lower IQ. The cognitive challenges include reduced EF, attention, and aspects of learning and memory due to deficits in the optimal roles of the prefrontal cortex (PFC), cerebellum and HPC. They will form the focus of the following sections, as well as a summary of the interconnectivity of these regions for children with DS.

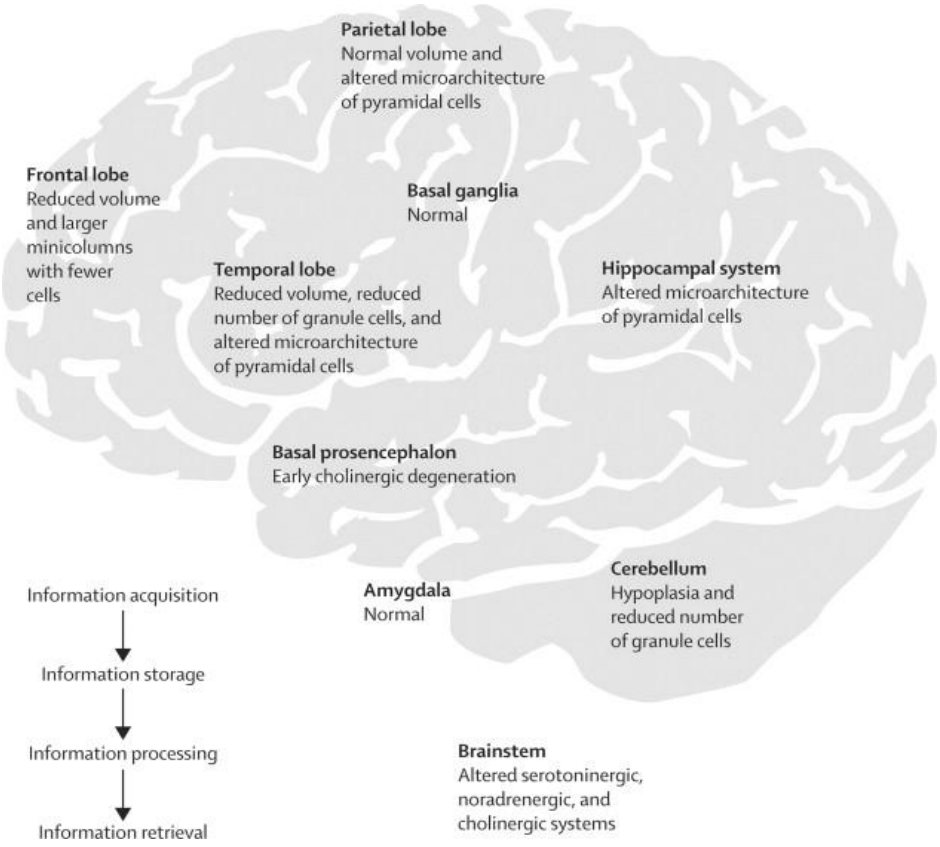


Figure 1.6. Cognitive deficits and associated neurological complications in individuals with Down Syndrome (Lott & Dierssen, 2010). I Lott and M Dierssen, 2010. Cognitive deficits and associated neurological complications in individuals with Down's syndrome, *Lancet Neural 9*(6), 623–633, Copyright 2024 with permission from *Elsevier*.

1.5 Structural and Functional Organisation in the Prefrontal Cortex

The PFC is located in the brain's frontal lobes, the largest cortical area of the brain, taking up 30% of the entire cerebral cortex (Carlén, 2017). It consists of the superior and medial frontal gyrus and the anterior part of the cingulate gyrus (Imada et al., 2021). The PFC lies anterior to the motor cortex, accounting for 10% of the brain. The PFC is not fully developed until a person's mid-20s but can be subdivided into the following regions: the Lateral PFC (including the Dorsolateral PFC (dLPFC) and Ventrolateral PFC (vLPFC), the Dorsomedial PFC (dMPFC), and the Ventromedial PFC (vMPFC or Orbitofrontal PFC (oFPFC) (Vaidya & Fellows, 2020).

The PFC processes and controls information from the cortical and subcortical areas, specifically the amygdala and the HPC. It is responsible for personality and behaviour and has a role in many vital functions, such as memory, attention, and working memory (WM) (Lara & Wallis, 2015). It also has a role in learning from mistakes, common sense, and executing decisions. Hence, it is the hub of EFs. Many of these functions are vital for what is considered a typical existence. Executive function, also called cognitive control, is essential for school, employment, mental and physical well-being, and quality of life (Diamond, 2012; Diamond, 2013).

The dLPFC is associated with attention. It allows someone to focus on a specific task and block out interference. Found at Brodmann area 46, this section of the PFC permits the WM to function. The orbital PFC processes emotion, Brodmann area 11, while the vLPFC is associated with empathy. The medial PFC is associated primarily with higher cognitive functions. Phineas Gage, for example, was a railroad worker in the mid-1800s. Gage was involved in an accident that resulted in a metal rod protruding through their skull, destroying most of their frontal lobe and PFC (O'Driscoll & Leach, 1998). It was believed that before this incident, Gage was diligent, hardworking and had a good temperament. However, their personality was altered after the incident, and they could not work well or make decisions, hence the significant role of the PFC in EF (O'Driscoll & Leach, 1998).

Initially, the EF was recognised as having two specific ways of processing information, both automatic and controlled (Schneider & Shiffrin, 1977). Hughes (2011) and Low and Simpson (2012) believe EF to be an overarching term that includes the complexity of cognitive abilities that can adapt to new contexts depending on the aim of the action (Low & Simpson, 2012; Amadó, Serrat et al., 2016). The most recent research has seen the division of the EFs to include ‘hot’ and ‘cold’. Hot EF is concerned with reward and emotion, whereas cold EF is associated more with higher-level processing cognitive tasks (Salehinejad, Ghanavati et al., 2021). This is believed to be underpinned by the vLPFC, vMPFC, and the orbitofrontal cortex while linked to the limbic system and related subcortical areas (Daunhauer, Fidler et al., 2014; Hongwanishkul, Happaney et al., 2005).

Cold EFs are associated with WM, attention, and problem-solving. The brain structures involved include the dLPFC, the lateral PFC, and the inferior frontal cortex,

which are linked to the subcortical areas, the HPC, and basal ganglia. These more logical processes will form the focus of what follows (Chan, Shum et al., 2008).

1.5.1 The Role of Fluid Intelligence

Diamond separates EF into three distinct areas dominated by a 'top-down mental process' when it is required to concentrate (Diamond, 2013). These include inhibition [inhibitory control, including self-control (behavioural inhibition) and interference control (selective attention and cognitive inhibition)] WM), and cognitive flexibility (Diamond, 2013). The WM is associated with the dLPFC, cognitive flexibility with the vLPFC, and inhibitory control with the vMPFC, which may be seen in Figure 1.7.

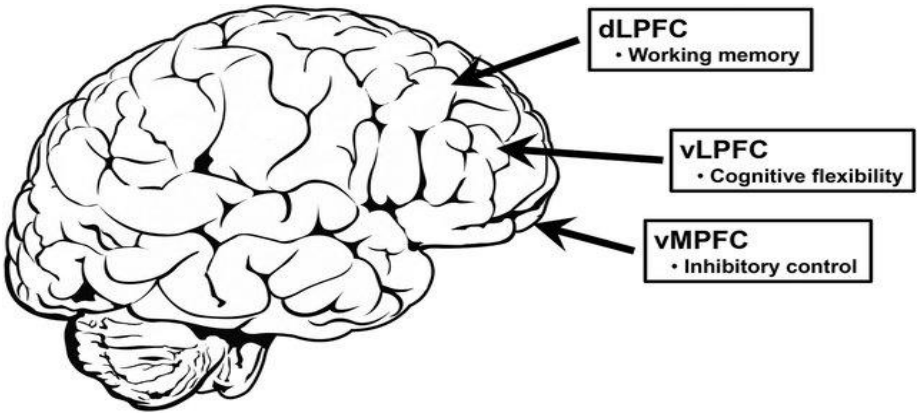


Figure 1.7. The dLPFC is associated with WM, the vLPFC with cognitive flexibility, and the vMPFC with inhibitory Control (Perry et al., 2018). R. Perry, E. Finegood, S. Braren & C. Blair (2018). The social neuroendocrinology and development of executive functions. In In book: Routledge International Handbook of Social Neuroendocrinology, 531–543. Reproduced with permission from *Taylor & Francis Books* (Creative Commons Attribution 3.0 License).

Inhibitory control is linked to controlling attention, behaviour, thoughts, and emotions regardless of external influences and doing what is more appropriate (Diamond, 2013). Inhibitory control of attention lets one focus on one thing while ignoring another stimulus (Diamond, 2013). Our attention can be stimulus-driven, bottom-up driven, or intentional, top-down dominated, referred to as executive attention (Posner & DiGirolamo, 1998; Theeuwes, 2010; Diamond, 2013). In addition, several types of inhibitory control, such as attention and action, share similar neural regions (Bunge, Dudukovic et al., 2002; Cohen, 2012; Diamond, 2013). Inhibitory control overlaps significantly with self-regulation, which is the ability to control emotional responses (Mischel & Ayduk, 2002; Eisenberg, Spinrad et al., 2010).

Working memory is vital for functioning within everyday society and relies on the dLPFC (Diamond, 2013). This kind of memory refers to a mental workspace, enabling a person to hold information in mind while simultaneously performing other complex cognitive tasks (Holmes & Adams, 2006). The WM is used in endless tasks, including reorganising lists and adding new information to existing information. Working memory and inhibitory control are interdependent (Diamond, 2013). For example, one holds information in mind and processes it to influence behaviour. Inhibitory control can prevent the WM from getting too cluttered by not giving unnecessary attention to information (Diamond, 2013). While both WM and inhibitory control are interlinked, it is still possible to study them separately. The prefrontal-parietal system supports WM, which allows us to remain focused on the material held in the mind without considering other irrelevant stimuli (LaBar, Gitelman et al., 1999; Awh, Anllo-Vento et al., 2000; Awh & Jonides, 2001; Ikkai & Curtis, 2011; Nobre & Stokes, 2011; Gazzaley & Nobre, 2012). However, as outlined by Davidson, young children are disproportionately challenged by inhibition control compared to adults (Davidson, Amso et al., 2006).

The terms WM and short-term memory (STM) are often used interchangeably. However, the difference between STM and WM is quite distinct when examined. In STM, one holds information while WM processes the data. For example, restating a parade of figures or objects in the order they are presented requires STM only, as this is just an exercise in repetition. However, if requested to repeat these figures in an alternative sequence, this would require WM. This temporary working space allows information to be processed and used in other cognitive areas (Baddeley & Hitch, 1974).

The central executive system (CES) is the 'boss' or the control system, coordinating the contrasting components of WM (Goswami, 2000). The CES is the WM manager that brings certain information to specific areas, as seen in Figure 1.8. The phonological loop deals explicitly with written and spoken communication, an integral component of the WM. The phonological loop stores and retains spoken language for a few seconds, while the articulatory control is connected to speech.

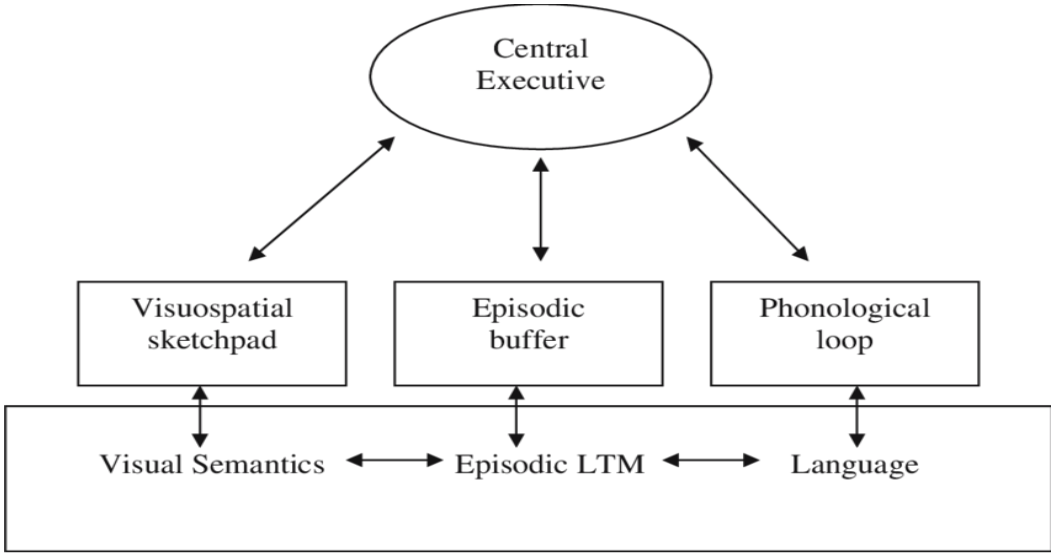


Figure 1.8. The revised working memory model includes an episodic buffer (Baddeley, 2000). A Baddeley (2000). The episodic buffer: a new component of working memory? *Trends in Cognitive Sciences*, *4*(11), 417–423. https://doi.org/10.1016/S1364-6613(00)01538-2. Reproduced with permission from *Elsevier*.

Some developmental psychologists have proposed that children's phonological awareness develops with age in accordance with increased vocabulary (Goswami, 2000). Initially, young children do not become aware of the phonetic features of a word but instead view the word holistically. However, as children get older, they begin to recognise the syllables and phonemes of a word. Their ability to segment words also coincides with increased WM capacities (Goswami, 2000). Goswami states that the number of items that can be held in the phonological loop measures a person's memory capacity (Goswami, 2000). The extent of this memory span varies depending on the material—for example, long words versus short words, etc. Therefore, digits are often used to test the memory span as they seem more accessible to everyone (Goswami, 2000). George Miller is one of the founding members of cognitive psychology and cognitive neuroscience. In 1956, he presented a paper called 'The Magical Number Seven, Plus or Minus Two: Some Limits on our capacity for process information' (Miller, 1956). He believed that the number of pieces of material that can be retained in WM was seven plus or minus two. This not only included individual pieces of material but also chunked material. Miller's law influenced the construction of phone numbers and teaching methodologies but was challenged in 2001 by Nelson Cowan (Norris & Kalm, 2021). Cowan re-examined

Miller's proposal, suggesting that the number of pieces of material that could be retained was four-plus or minus two (Cowan, 2010).

Speech rate is also an essential element that affects memory span (Goswami, 2000). It is believed that children who articulate at a slower pace have a shorter memory span than those who communicate quicker (Hulme, Thomson et al., 1984; Burkholder & Pisoni, 2003). This was also proven to be the case in a study by Leonard et al., who tested 14 children with language impairment (LI) and their TD peers (Leonard, Weismer et al., 2007). The research team tested speed processing, WM, and visual working memory (VWM) through this study. The results showed that the children with LI did not perform as well on the tasks, with VWM being the most significant contributing factor (Leonard, Weismer et al., 2007). Hulme thought that the faster a child could speak increased their capacity to rehearse and retain more information (Hulme, Thomson et al., 1984). Speech rate and memory span also depend on the 'phonological representations' (Henry & Millar, 1993). Growing evidence shows enhanced WM depends on items with strong phonological representation in long-term memory (LTM). Therefore, it is more accessible to reintegrate information, leading to improved WM (Goswami, 2000).

The visuospatial sketchpad is responsible for visual and spatial elements in WM (Goswami, 2000). It is considered the 'inner eye' of the WM and is crucial in how we view ourselves concerning other objects and how we move within our environment. Any visual distraction can impair the performance of the sketchpad. As we move around, information is continually changing, and we need to keep track of these constantly evolving situations. Just as there is a relationship between sensory and motor skills in speech, there is also a similar relationship in visuospatial memory. Saccadic eye movements are as vital in maintaining spatial locations as articulatory rehearsal is for maintaining verbal memory (Postle, 2006). Interestingly, these visual and verbal systems can operate at the same time. Baddeley added the episodic buffer, a backup memory that communicates between LTM and WM. The episodic buffer brings information from the phonological loop and the visuospatial sketchpad to create a unified memory (Henry et al., 2010).

Diamond identifies cognitive flexibility as being important in EF. Cognitive flexibility involves finding contrasting approaches to a problem or alternating

between tasks (Diamond, 2013). It is also the ability to 'think outside of the box', which is why it is present later in development, usually from 12 years into adulthood (Davidson, Amso et al., 2006; Garon, Bryson et al., 2008). There is a significant link between creativity and cognitive flexibility (Diamond, 2013).

All previously discussed aspects of 'higher-level executive functions' are responsible for reasoning, problem-solving, and planning without previous knowledge. When these are done in real time, it creates 'fluid intelligence' (Diamond, 2013). Problem-solving, planning, and reasoning are synonymous with fluid intelligence (Ferrer, O'Hare et al., 2009). Figure 1.9 provides a detailed diagram of how WM, inhibitory control, and cognitive control work together to create higher executive functions.

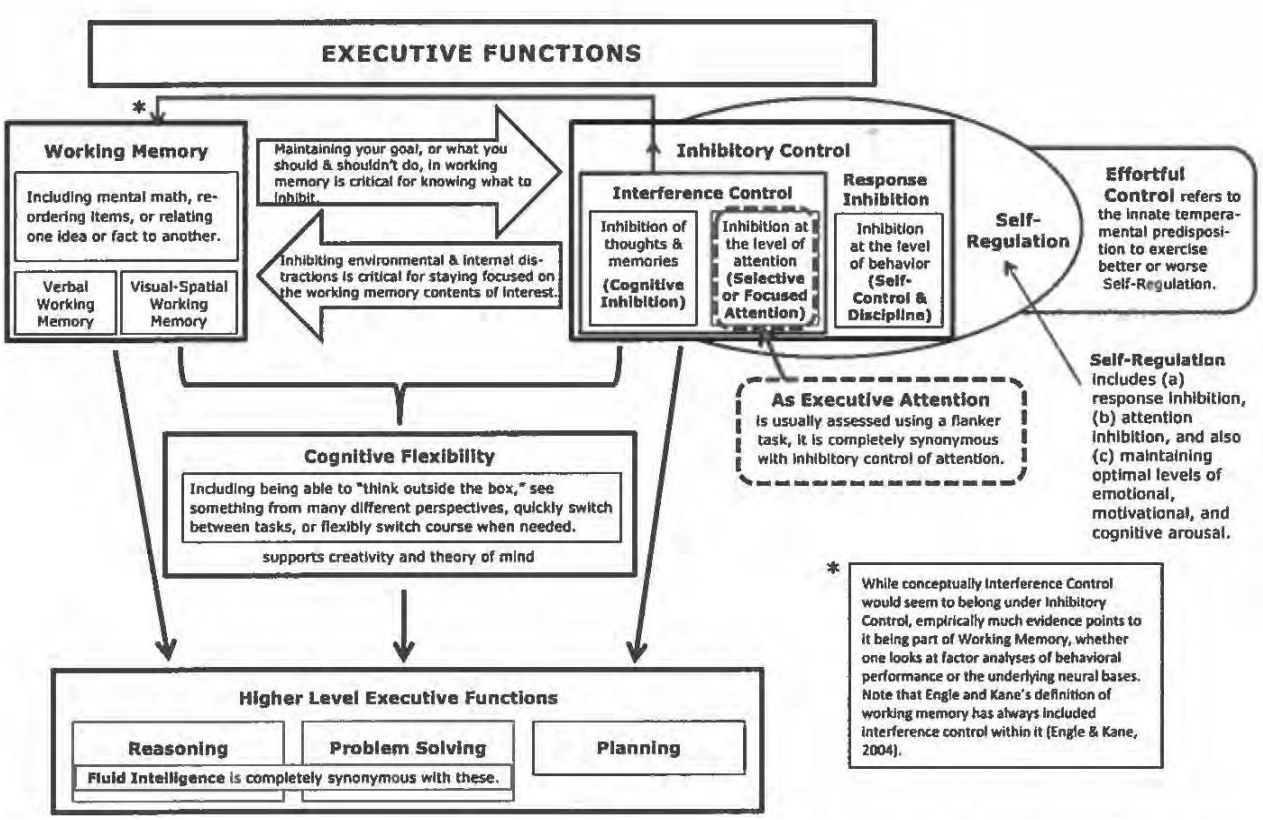


Figure 1.9. Executive Functions and related terms (Diamond, 2013). A. Diamond (2013). Executive Functions. *Annual Review of Psychology*, *64*(1), 135–168. https://doi.org/10.1146/annurev-psych-113011-143750. Reproduced with permission from *Annual Reviews*.

1.5.2 Prefrontal Cortex Structure and Function in Those with Down Syndrome

The frontal lobes in the brains of children with DS are disproportionally smaller than their TD counterparts (Nadel, 2003). As the PFC is considered a late-developing neural system, the alteration in cognitive function becomes increasingly evident throughout the life span of a person with DS (Edgin, 2013). Nadel's earlier research believes the syndrome might be defined by these late-developing neural systems, including the frontal lobes, the medial temporal lobe (MTL), and the cerebellum. While some morphological changes occur prenatally, the most rigorous alterations to some brain regions occur postnatally. One study assessed brain development in 101 children between birth and five years (Wisniewski, 1990). This study found that the DS brain was of equal size and shape to the TD brain at birth. However, even within the first 3–5 months, the anterior-posterior (A-P) diameter was shorter and secondary to the reduction in the frontal lobes, which were 22.5% smaller in the DS cohort compared to 6.8% in TD children (Wisniewski, 1990). The occipital poles were also flatter, while the cerebellum and brain stem in the DS cohort were smaller (a, c) than those in the TD group (b, d), as seen in Figure 1.10.

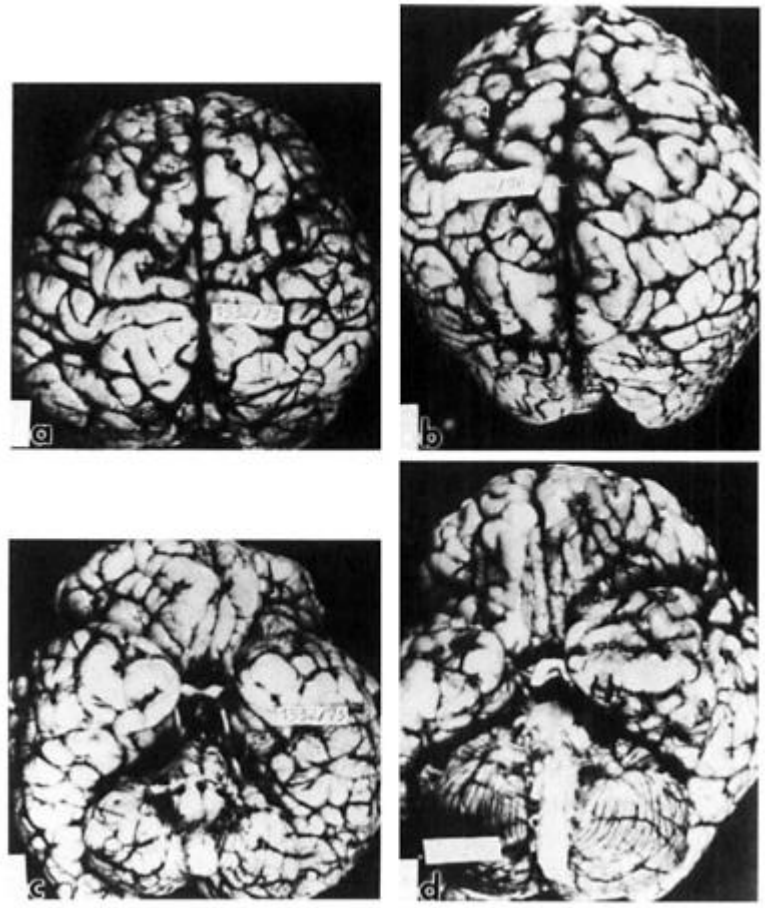


Figure 1.10. The anterior-posterior (A-P) diameter was shorter and secondary to the reduction in the frontal lobes in the DS cohort compared to the TD children. The occipital poles were also flatter, while the cerebellum and brain stem in the DS cohort were smaller (a, c) than those in the TD group (Wisniewski, 1990). K. E Wisniewski (1990). Down syndrome children often have a brain with maturation delay, retardation of growth, and cortical dysgenesis. *American Journal of Medical Genetics*, *37*(57), 274–281. https://doi.org/10.1002/ajmg.1320370755. Reproduced with permission from *John Wiley & Sons INC*. (Blackwell Publishing Ltd.).

In a recent study, Lee and colleagues used DTI-driven tensor-based morphometry (DTBM) to assess the levels of hypoplasia in the brains of school-going children with DS. They found significant hypoplasia in the frontal lobes (Lee, Nayak et al., 2020). This reduction in cells in the frontal lobes has an adverse effect on the PFC of those with DS and hence impacts the EFs of this cohort.

As addressed previously, EF refers to top-down processes that include attention, WM, planning reasoning, and impulse control. These are required to accomplish goal-directed behaviour and are vital in everyday life (Daunhauer, Fidler et al., 2014). Children aged between 2 and 4 explicitly share delays in these cold EFs. Studies have shown that toddlers with DS show limited problem-solving strategies when retrieving an object (Daunhauer, Fidler et al., 2014). The authors believe these

strategies are the first form of nonverbal communication that could impact developments in other areas, such as language (Daunhauer, Fidler et al., 2014). One study carried out by Daunhauer et al. explored the 'Profiles of everyday executive functioning in young children with Down syndrome' by comparing the EF of those with DS (n=25) with MA-matched TD children (n=23). The researchers acquired both teacher and parent reports wherein they asked three questions:

1. Do young children with DS present with significant levels of deficits in everyday EFs compared to the MA-matched comparison group of TD children as reported by (a) teachers and (b) parents?
2. Do teachers concur with their reports of everyday EF function in children with DS?
3. What is the relative risk for school-age children with DS presenting with executive dysfunction as reported by (a) teachers and (b) parents? (Daunhauer, Fidler et al., 2014).

The Behaviour Rating Inventory of Executive Function-Preschool (BRIEF-P) test assessed children's EF at preschool age. As noted by the teachers, the DS cohort experienced impairments in WM (68.4%) and planning (47.4%) (Daunhauer, Fidler et al., 2014). The parents detected that 64% had deficits in WM, 40% in planning, and 32% in inhibitory control. It must be acknowledged that the BRIEF-P does not account for cold and hot EF. However, as Daunhauer et al. suggest, the Index Emergent Metacognition has scales for WM and planning, as has previously been identified as these cold domains. The implications of these areas of EF for those with DS will be assessed in the following section.

Several studies have detailed the challenges in the WM of memory in children with DS. In the report above by Daunhauer et al., children with DS were nine times more likely to experience WM challenges at home or school. However, visual short-term memory (VSTM) and WM are more affected in those with DS than visuospatial memory, which is believed to be a particular strength in this cohort.

Challenges with language, communication, and speech are some of the prevailing characteristics of DS (Martin, Klusek et al., 2009). However, a wide range of individual differences exist in children with DS concerning language acquisition. Zampini and D'Odorico conducted a study to explore the link between gesture and vocabulary development in toddlers with DS (Zampini & D'Odorico, 2008). Twenty children with DS were initially assessed during a play session they attended with their mothers. While their psychomotor development was assessed using the Brunet-

Lézine Scale of Infant Development, their vocabulary, both expression and comprehension, was evaluated using the MacArthur Communicative Development Inventory (CDI). When tested, they found that the lowest score was in non-verbal children, whereas the highest score was in a child that produced 234 words. Parents were then asked to complete a questionnaire six months after the initial evaluation. The researchers then analysed the relationship between gesture production, vocabulary size and psychomotor development. After this time, the non-verbal child remained unable to communicate verbally, while the high-scoring children had 500 words (Zampini & D'Odorico, 2008). In conclusion, they found that gesture production at 36 months was significantly related to subsequent vocabulary production in this cohort (Zampini & D'Odorico, 2008).

Deckers et al. conducted a study with thirty children between 2 and 7 years old to establish a core vocabulary list for those with DS. Deckers et al. found that 50 core words made up 67.2% of all words used by the cohort, and therefore, this could assist Speech and Language Therapists (SLTs) in addressing focused language in this population (Deckers, Hof et al. 2017). Parents and professionals use the Down Syndrome Education (DSE) checklists to assess children's vocabulary and communication skills.

While there is substantial individual variability of strengths in this area, receptive language is more robust in this cohort than expressive language, phonology, and syntax (Fowler, 1990; Chapman, Seung et al., 1998; Martin, Klusek et al., 2009; Næss, Lyster et al., 2011). Syntax poses considerable difficulty for children and adults with DS (Frizelle, Thompson et al., 2018). Research has shown that while many children with DS have hearing and speech challenges, it does not account for difficulties with VSTM (Jarrold & Baddeley, 2001; Laws, 2002; Martin, Klusek et al., 2009). Therefore, researchers have spent much time unravelling the underlying causes of these challenges in the VSTM, including the most recent study by Penke and Wimmer that explored the link between VSTM deficits and sentence comprehension (Penke & Wimmer, 2020). While at first it was believed that the VSTM hindered language development in this cohort (Gathercole & Baddeley, 1990; Laws & Gunn, 2004; Pennington et al., 2010), it is now believed to be specific to the phonological loop rather than the semantic contributions to VSTM (Hulme &

Mackenzie, 1992; Buckley, Broadley et al., 1995; Das & Mishra, 1995; Comblain, 1996; Jarrold & Baddeley, 1997; Lee et al., 2004). The phonological loop is fundamental to language learning, reading, and vocabulary development (Baddeley, Gathercole et al., 1998; Buckley & Bird, 1998). The loop includes speech input, subvocal rehearsal and the phonological store.

Discrepancies in VSTM in this cohort are due to memory capacity limitation in the phonological store. Children with DS have an issue with the executive load (McDade & Adler, 1980; Lanfranchi, Cornoldi et al., 2004). Chapman et al. studied 47 participants with DS and compared their memory span with TD cohorts (Chapman, Seung et al., 1998). The average result amongst the TD cohort was between 3–6 items, with the typical being five. Within the DS group, while one achieved six items, the standard was 4, with a couple reaching just 2. The store is passive; therefore, it is only through 'subvocal' rehearsal, as in repeating the material to oneself, that the material can be retained. This is believed to occur in TD children from 7 years (Baddeley, Gathercole et al., 1998). However, a study by Varnhagen et al. found that when those with DS were asked to remember four items in a phonological similarity effect test, the DS participants had only a span of two objects (Varnhagen, Das et al., 1987). Therefore, the list of four things exceeded their STM memory capacity. Consequently, all phonological coding is abandoned (Hall, Wilson et al., 1983; Salamé & Baddeley, 1986; Gathercole & Baddeley, 1990; Varnhagen, McCallum et al., 1997).

The VSTM relates to how children say the words and their length (Baddeley, Thomson et al., 1975). Rehearsing is difficult for those with DS as they do not repeat effectively and rely on the phonological store, which is a passive entity (Hulme & Mackenzie, 1992). Therefore, store, rehearsal, and phonology account for the limitations in this VSTM in children with DS (Hulme & Mackenzie, 1992; Broadley, 1993; Comblain, 1996). Their lack of ability to articulate the lists at a quicker pace unavoidably affects their VSTM (Cowan, Day et al., 1992).

Unsurprisingly, interventions have focused on enhancing memory skills in those with DS through rehearsing training to increase verbal span (Comblain, 1999; Conners, Rosenquist et al., 2008). The Sarah Duffen Centre has been using memorising and rehearsing games since 1990 (Buckley, Broadley et al., 1995; Laws, Taylor et al., 1996). In addition, Laws encouraged 25 children with DS to repeat a list

of items when a new item was added. It involved eighteen training sessions of 15-minute durations for six weeks, after which they found improvement in the tasks related to digit rehearsal (Laws, Taylor et al., 1996). While these studies have addressed this rehearsal method, it is now firmly believed that the deficits in phonological STM in individuals with DS originate in storage rather than either encoding or rehearsing (Broadley, 1993; Buckley, Broadley et al., 1994; Buckley, Broadley et al., 1995; Comblain, 1995; Laws, Taylor et al., 1996; Baddeley & Jarrold, 2007).

1.6 Cerebellum: Structure and Function: The Increasing Role of the Cerebro-Cerebellar Circuitry

Latin for 'little brain', the cerebellum is found at the base of the brain. It is the most extensive section of the hindbrain and is inferior and posterior to the cerebral cortex. It consists of two cerebellar hemispheres in three parts (Figure 1.11). The cerebrocerebellum receives information from the cerebral cortex regarding planning and initiating movements. The spinocerebellum receives information about limb position and pressures. The vermis cerebellum runs through the cerebellum and involves limb, posture, and eye movements. The vestiobulcerebellum is concerned with equilibrium, balance, and posture. It communicates with the rest of the brain through the cerebella peduncles, the superior, middle, and inferior peduncles. When information is sent to the cerebellum, it takes an extensive path to the Purkinje cells, which then project to the nuclei in the centre of the cerebellum, the deep cerebellar nuclei. Information is then sent to the brain stem and the thalamus. While it only constitutes 10% of the entire brain, it contains 80% of the total number of neurons found in the brain (Sathyanesan, Zhou et al., 2019). It also develops postnatally, and in the first 90 days, it increases in size by 108% (Sathyanesan, Zhou et al., 2019). The cerebellum is, therefore, vulnerable to many neurodevelopmental conditions.

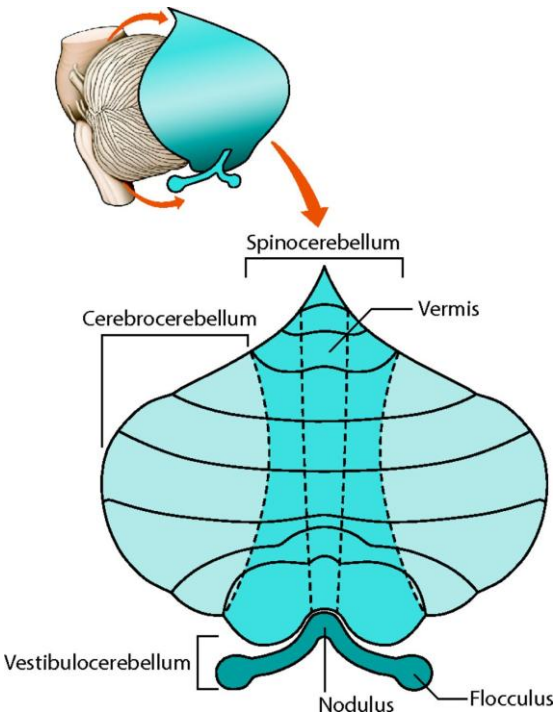


Figure 1.11. The cerebrocerebellum receives information from the cerebral cortex regarding planning and initiating movements. The spinocerebellum receives information about limb position and pressures. The vestiobulcerebellum is involved with equilibrium, balance, and posture. It knows where a limb is and if it needs to be corrected in space (Klein, Ulmer et al. 2016). A. P. Kelin, J. L Ulmer, S.A, Quinet, V. Mathews. (2016). Nonmotor Functions of the Cerebellum: An Introduction. *American Journal of Neuroradiology*, *37*(6), 1005. https://doi.org/10.3174/ajnr.A4720. Reproduced with permission from the *American Society of Neuroradiology*.

The importance of the cerebellum in higher-order cognition and emotions has received much attention in recent years (Bordignon, Devita et al., 2021). The first study published by Brogden and Gantt in 1942 explored the contribution of the cerebellum to learning and memory (Brogden & Gantt, 1942). The cerebellum creates non-declarative memories specific to skills, habits, and learning (Squire et al., 1994; Thomas & Kim, 1996). Raichle and colleagues found that the cerebellum impacted learning and word generation (Raichle, Fiez et al., 1994). Significant research by Peterson et al. identified that the cerebellum was recruited in a semantic association task that asked participants to select verbs to match the noun given (Peterson et al., 1998). It was seen that the lateral section of the cerebellum was activated during this task. This non-motor function of the cerebellum is not surprising due to its proximity to the cerebral cortex (Palesi, De Rinaldis et al., 2017). Additional connections are made to many other brain areas through the thalamus, which are responsible for behaviour and cognition, including the previously mentioned dLPFC. Evidence for alternate functions is also contextualised by the cerebro-cerebellar loop, where

connections go from the cerebral cortex to the cerebellum (Tanaka, Ishikawa et al., 2020). The cerebellum and the cerebral cortex are connected through two main pathways. The cerebello-thalamo-cortical (CTC) pathway and the afferent cortico-ponto-cerebellar (CPC) pathway are seen in Figure 1.12. The extent to which the cerebellum is involved in other functions through the cerebro-cerebellar circuitry is still unclear, yet further research may reveal valuable information about the influence of this brain region and how it develops throughout our life span.

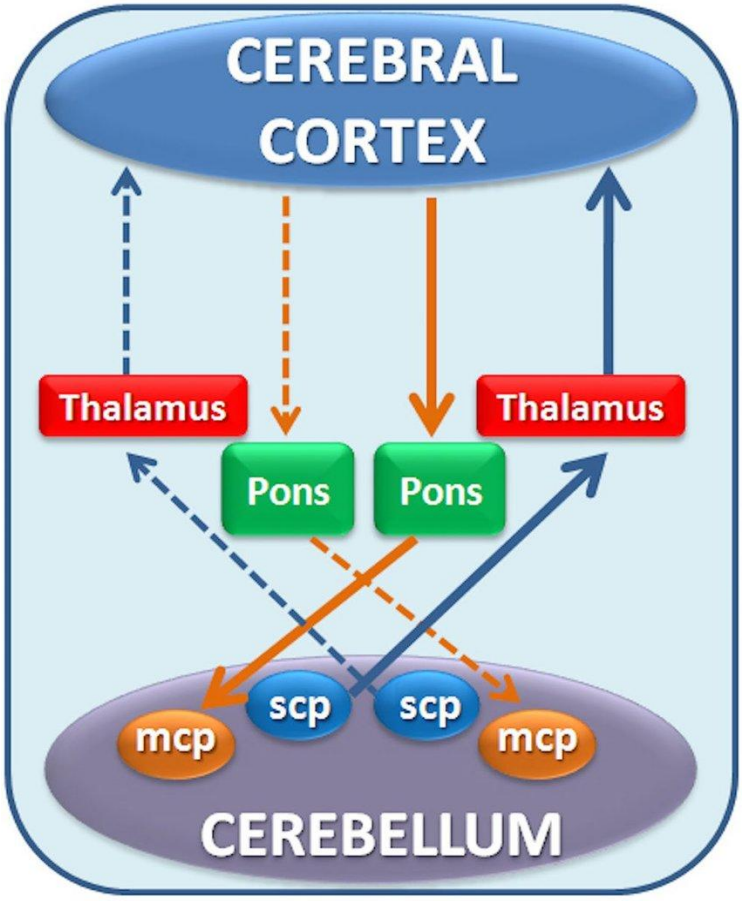


Figure 1.12 The cerebellum and cerebral cortex are connected through two main pathways: the cerebello-thalamo-cortical (CTC) and cortico-ponto-cerebellar (CPC) pathways. The orange arrows connect the cerebrum with the cerebellum through the pons and the middle cerebellar peduncle (MCP). The blue arrows connect the cerebrum through the superior cerebellar peduncle (SCP), called the cerebello-thalmo-cortical (CTC) pathway. The dotted arrows are contralateral pathways (Palesi, De Rinaldis et al., 2017). Palesi, Fulvia & Rinaldis, Andrea & Castellazzi, Gloria & Calamante, Fernando & Muhlert, Nils & Chard, Declan & Tournier, Jacques-Donald & Magenes, Giovanni & D'Angelo, Egidio & Gandini Wheeler-Kingshott, Claudia. (2017). Contralateral cortico-ponto-cerebellar pathways reconstruction in humans in vivo: Implications for reciprocal cerebro-cerebellar structural connectivity in motor and non-motor areas. Scientific Reports. 7, 2. https://doi.org/10.1038/s41598-017-13079-8. Reproduced with permission from *Springer Nature*. (Creative Commons Attribution 3.0 Licence).

1.6.1 Cerebellar Variation in Children with Down Syndrome

The cerebellum changes in size, structure, and volume pre- and postnatally. In TD populations, the cerebellum quadruples its volume between prenatal weeks 24 and 38, increasing its cortical surface thirty times (Volpe, 2009; Sathyanesan, Zhou et al., 2019). Therefore, much focus has been on the alterations to cerebellar development in the DS foetus prenatally in the second trimester to help pinpoint these developmental changes and when they occur (Patkee, Baburamani et al., 2020). Most recently, Patkee et al. conducted a study to track the cerebellar development of foetuses with DS in utero and postnatally against a control group of TD babies. The TD control group included 52 foetuses and 21 neonates, while 30 DS foetuses, 17 with a cardiac defect and 13 without, were included in the study. Six of the foetuses were then scanned as neonates. Differences in cerebellar volume occur in the second trimester, with a considerable decline in the vermis height and surface in the DS group. The transcerebellar diameter and vermis width appeared smaller in the third trimester.

Cerebellar hypoplasia is a significant factor in those with DS and accounts for the lack of muscle tone (Miguel, 2010). Lee et al. conducted a study using a scanning approach that extracts morphological information about hypoplasia in the cerebellum. They recruited 15 participants with DS and 15 age and sex-matched TD controls. All participants were between 6 and 24 years old, with a mean age of 17 (Lee, 2020). DTI-driven tensor-based morphometry (DTBM) revealed significant hypoplasia in the afferent pathways in the cerebellum. Specifically, the connections between the cerebellum's fronto-pontine (middle cerebellar peduncle) and olivo-cerebellar (inferior cerebellar peduncle) were affected. The cerebro-cerebellar circuits can help shape brain development (Stoodley, 2016); therefore, further investigation is needed to assess the impact of such on cerebro-cerebellar hypoplasia in this cohort. Evidence shows a shortening in dendritic length and cerebral cortical pyramidal cells with fewer synapses, resulting in shorter local connections rather than inter-hemispherical connections (Lott, 2012).

Both gross and fine motor skills in people with DS are compromised due to cerebellar alterations, with posture and balance mostly affected (Lee, Nayak et al.,

2020). A child begins to explore the world through reaching, grabbing, and crawling; however, children with DS encounter challenges at this stage of development (Malak, Kostiukow et al., 2015). This is due to the slower development trajectory of the cerebellum in this cohort, which becomes evident after six months as these children fail to reach their milestones compared to their peers. Children with DS also encounter difficulty standing. In a study carried out by Malak et al., 95% of children with DS could stand upright between the ages of 3 and 6, with 10% being able to stand before three years of age, whereas TD children begin to stand between 9–10 months (Malak, Kostiukow et al., 2015). The development of the ability to stand is also delayed by muscle hypotonia, which affects these children. Therefore, acquiring the skills of walking and running is additionally influenced. Developing these motor skills at a slower rate thus impacts social, cognitive, and further motor skills.

As stated by Memisevic and Macak, there are two principles of development: cephalous-caudal and proximal-distal (Memisevic & Macak, 2014). The proximal-distal principle concerns trunk strength and central access control necessary for hand usage (Memisevic & Macak, 2014). Therefore, the development of fine motor skills in children with DS depends on gross motor skills. If the latter is already delayed, this similarly affects the developmental trajectory of fine motor skills required for everyday life activities such as dressing, feeding, and holding objects such as pencils (Memisevic & Macak, 2014). Significant research has confirmed a link between fine motor skills, academic achievement, and social, cognitive, and emotional development (De Luca, McCarthy et al., 2013). Gligorović et al. conducted a significant research study which showed a positive correlation between motor skills and academic achievement (Gligorović & Durovic, 2012). Consequently, children with DS are academically impeded because of delayed motor skills development due to cerebellar aberrations.

Research has been carried out since the 1980s that confirms the cerebellum has a role in non-motor functions such as cognition and emotion (Turner, Paradiso et al., 2007; Sathyanesan, Zhou et al., 2019). Additional studies have connected the cerebellum to sensory processing, including the auditory, visual, tactile, and olfactory processes (Deluca, Golzar et al., 2014; Baumann, Borra et al., 2015; Sathyanesan, Zhou et al., 2019). It has been well-documented that people with DS have hearing

difficulties and encounter sensory problems. Therefore, the dramatically reduced volume in the cerebellum could account for some of this cohort's intellectual and linguistic challenges.

1.7 Hippocampal Structure in the Typically Developing Population

The hippocampus (HPC) (Greek: Seahorse) is located in the temporal lobe of each cerebral hemisphere (left and right). It comprises the c*ornu ammonis* (CA) 1–4, the dentate gyrus (DG), and the subiculum. Figure 1.13 shows a detailed dorsal view of the HPC with the DG inside.

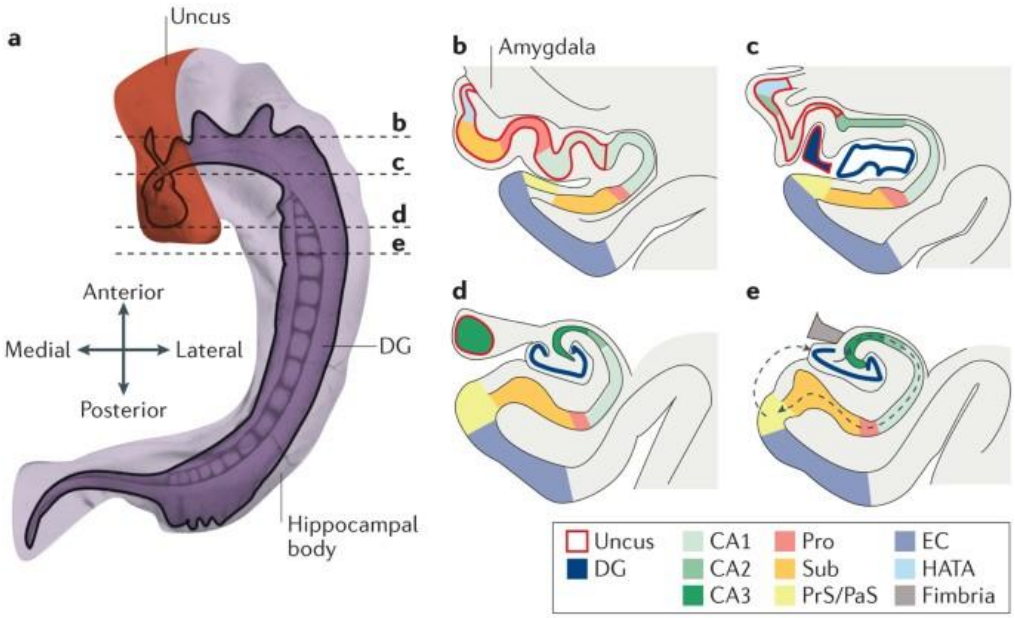


Figure 1.13. This is the anatomy of the anterior hippocampus; it shows a dorsal view of the hippocampus with the dentate gyrus (DG). The main body and the uncus may be seen in b–e. The arrow in e indicates the flow of information in the hippocampal circuity from the entorhinal cortex (EC) to the hippocampal-amygdaloid (HATA) area to the pro-subiculum to the presubiculum (PrS) and the parasubiculum (PaS). P. Zeidman, & E. A. Maguire, (2016). Anterior hippocampus: the anatomy of perception, imagination and episodic memory. In *Nat Rev Neurosci*, *17*(3), 173–182. https://doi.org/10.1038/nrn.2015.24. Reproduced with permission from *Springer Nature*.

The HPC has three layers: an outer molecular layer, a middle pyramidal layer, and an inner polymorphic layer. CA4 is not part of the c*ornu ammonis* and is, in fact, the dentate *hilus* (RajMohan & Mohandas, 2007). *Cornu Ammonis* was named and identified by Lorente de Nó in 1934. The dentate *hilus* is another name for the polymorphic layer, C4, the junction between the HPC and DG. The fornix is the primary output region for the HPC and connects to the mammillary bodies, while the

CA1 joins the subiculum to the HPC proper. The evident neurogenesis (birth of new neurons) in this brain region throughout adulthood supports the idea that the brain, regardless of age, is receptive to new experiences that can help change behaviours, hence the role of the HPC in brain plasticity (Maguire, Gadian et al., 2000).

The internal circuit of the HPC receives fibres from the entorhinal area, DG, *cornu ammonis*, and subiculum. Figure 1.14 shows three pathways in this area: the perforant pathway, mossy fibres, and Schaffer collaterals, collectively called the trisynaptic loop. The perforant pathway is considered the predominant pathway to the HPC. The fibres from the entorhinal site 'perforate' the subiculum, reaching the DG, crossing the HPC fissure to the CA3 and then to the fimbria. CA3 then links the Schaeffer collaterals that extend to the dendrite of CA1, which is the main output of the HPC that similarly extends to the alveus, fimbria, and fornix (RajMohan & Mohandas, 2007). The DG consists of three layers: the outer acellular molecular layer, the granular middle layer, and an inner polymorphic layer (RajMohan & Mohandas, 2007). The function of the DG is to carry information from the entorhinal cortex (EC) to CA3 via the perforant pathway (Puelles, 2009; Yuste, 2015).

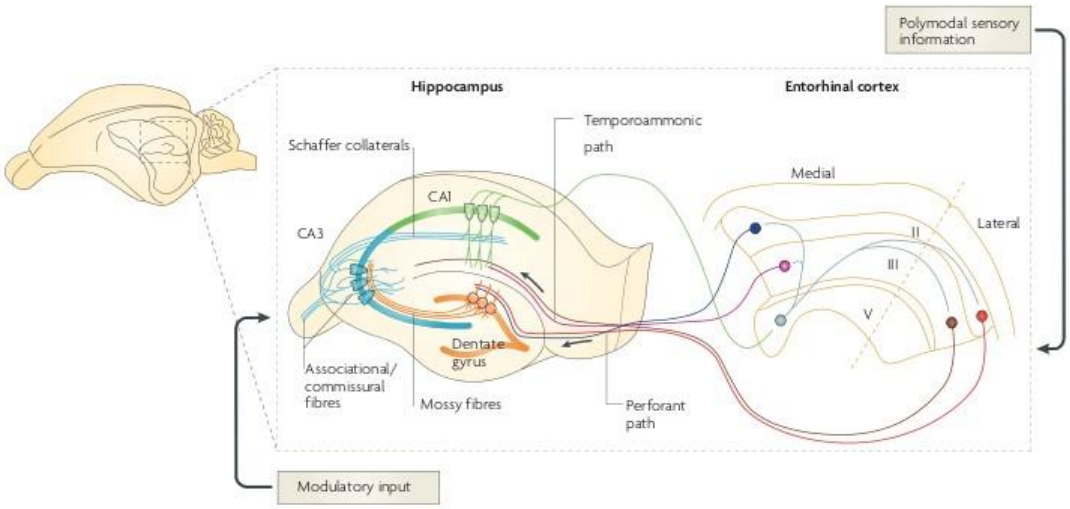


Figure 1.14. The trisynaptic loop (Küffner, 2013.) M. Küffner (2013). Ultrastructural analysis of spineapparatus in CA3 pyramidal neurons following single-cell electroporation in Synaptopodin Knockout mice. Reproduced with permission from *Elsevier*. (Creative Commons Attribution 3.0 License).

1.7.1 Hippocampal Function in Typically Developing Children

Volumetric magnetic resonance imaging (MRI) has proven that maximum growth of the HPC in TD children occurs in the first two months after birth, with additional rapid growth up to the age of two years (Knickmeyer, Gouttard et al., 2008; Thompson, Omizzolo et al., 2014). This growth rate usually peaks between 9 and 11 years of age and then slows down (Braun et al. et al., 2014; Thompson, Omizzolo et al., 2014). The HPC is primarily responsible for learning, memory, and spatial navigation and plays a vital role in early childhood development (O'Keefe & Nadel, 1978). In many ways, learning and memory are interdependent. Learning is how we acquire knowledge, whereas memory processes the information, encodes it, and recalls it (Kandel, 2000). The functions of neural circuitry are hugely influential in education. This reflects the neuroscientific theory 'Hebb's Rule' that claims that neurons adapt during the learning process and hence 'neurons that fire together wire together' (Hebb, 1949). An action potential occurs when the electrochemical wave travels along the axon of a neuron. This then releases a neurotransmitter that helps connect neurons across the synaptic cleft. Also known as the neuronal junction, a synapse is a space between two neurons. This gap allows the neurons to transmit information. Most synapses are chemical, where the presynaptic neuron fires a neurotransmitter. However, those that are electrical send ions to the next neuron. An axon can have several branches that can allow several synaptic connections to be made. Similarly, a neuron can receive several synaptic messages, as seen in Figure 1.15. Some synaptic vesicles in the axon terminal can fill with neurotransmitter molecules. The terminal and the pre- and postsynaptic cell gaps are called the synaptic cleft.

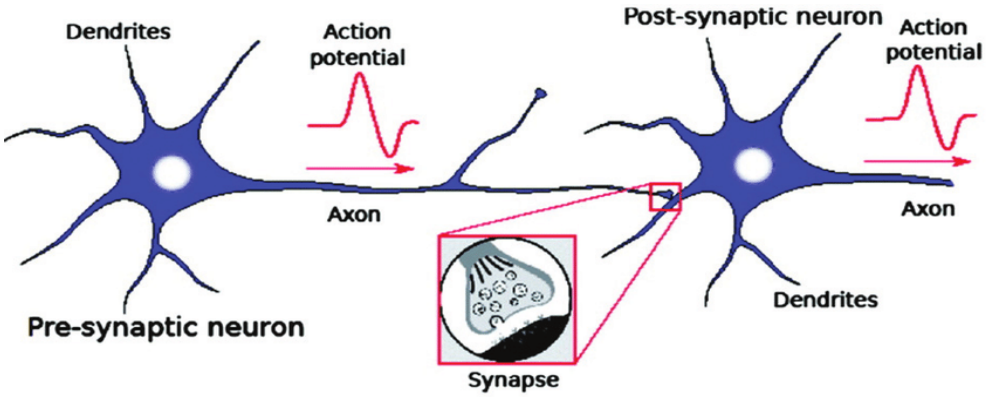


Figure 1.15. Neuron and Synapse (Anping, Xinjiang et al., 2017). Memristor Neural Network Design. In J. Alex Pappachen (Ed.), *Memristor and Memristive Neural Networks*, Ch. 12). *IntechOpen*. https://doi.org/10.5772/intechopen.69929. Reproduced with permission from *IntechOpen*. (Creative Commons Attribution 3.0 License).

Synaptic plasticity and cognitive processes concerning the HPC have been extensively researched (Cramer & Galdzicki, 2012). Synaptic plasticity dictates how effectively two neurons communicate with each other. Through such communication, the structures and functions of the synapses become altered in response (Cramer & Galdzicki, 2012). Both pre- and postsynaptic activity can weaken or strengthen learning and memory potential (Cramer & Galdzicki, 2012). This can lead to short-term or long-term synaptic plasticity; the latter influences how we store and access memories. This can result in dendritic arborisation (Carr & O'Mahony, 2019). The dendrites grow out of 'activity-dependent plasticity'; the more often they are used, the more they grow, supporting enhanced memory and learning. An excellent analogy of the function of the HPC is found in the publication *Brain Centric Design*, in which Carr and O'Mahony compare the HPC to a search engine. A chapter entitled 'Google is like your Hippocampus' describes how it is comparable to how Google operates. When a webpage is created, Google indexes it, and when someone searches keywords, it is retrieved. The web pages retrieved most often are prioritised, and those not accessed as often are repressed (Carr & O'Mahony, 2019).

Long-term potentiation (LTP) is a process whereby the synaptic connections between neurons become strengthened through increased activation. Therefore, LTP is a cellular process underlying memory and learning. This formation of synaptic connections is most active in early childhood, and the more certain experiences occur, the more connections become robust; consequently, the more experiences and skills that are not revisited are pruned ('use it or lose it').

Substantial research has been conducted on the HPC, particularly its role in memory, learning, and emotion (Opitz, 2014; Richter-Levin, 2004). The ventral hippocampus (VH) is primarily associated with emotions linked to the limbic system (Bannerman, Rawlins et al., 2004). 'Limbic' comes from the Latin *Limbus,* meaning border. It is a border between the cerebral cortex and the subcortical structure of the diencephalon. Paul Pierre Broca, in 1878, first applied the term limbic to the curved rim of the cortex, speaking of 'le grand lobe limbique' (RajMohan & Mohandas, 2007). However, it was Paul D. MacLean who coined the term 'limbic system' to describe Broca's limbic lobe, which he named in 1952, including the cingulate and parahippocampal gyri (Nakano, 1998). The limbic system also includes the HPC, DG, amygdala, hypothalamus, septal area, and thalamus (Esch & Stefano, 2005).

The HPC is a principle area linked to regulating response to stressors. The HPC is directly related to the hypothalamus-pituitary adrenals, hence the HPA axis. When a stressor is introduced and communicated through the amygdala and the HPC, the HPA produces corticotropin-releasing hormone (CRH). This causes the pituitary gland to produce adrenocorticotropic hormone (ACTH), producing glucocorticoids, of which cortisol is the main one produced by the adrenal gland, as seen in Figure 1.16. When this happens, negative feedback is sent to the HPC to prevent too much cortisol from being produced when the stressor ceases (Jacobson & Sapolsky, 1991). According to Lee, the VH has also been associated with WM concerning odour (Lee, Lew et al., 2019).

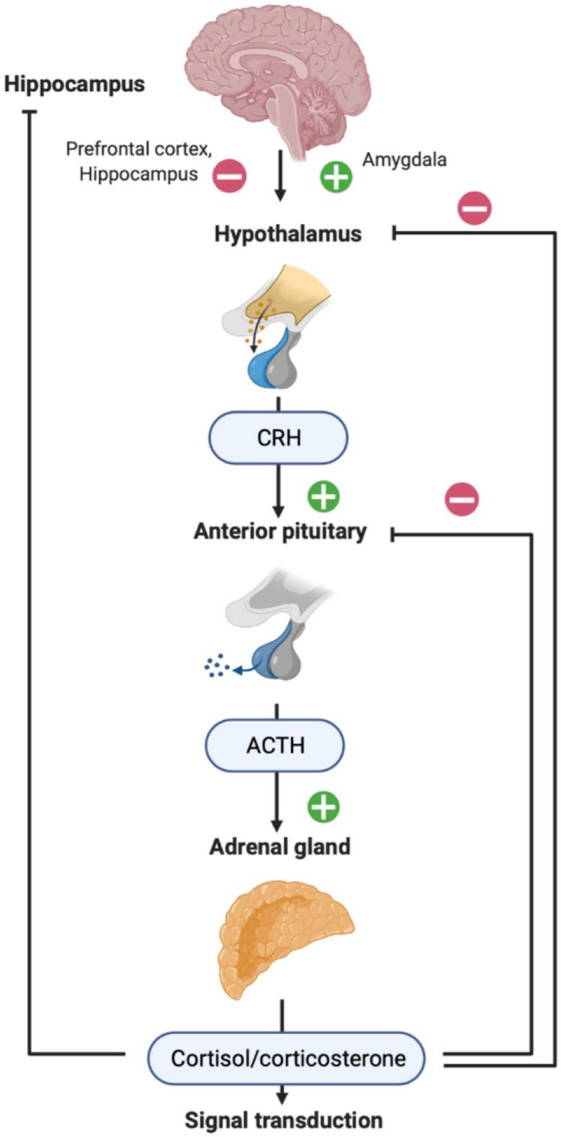


Figure 1.16. The schematic connection between the hippocampus and the hypothalamus-pituitary-adrenals (HPA) axis. As seen here, stress triggers the release of glucocorticoids, which then feed back to the hippocampus, hypothalamus, and pituitary. Acetylcholine (ACh) is also involved in facilitating neuroendocrine, emotional, and physical responses in relation to the HPA axis.(Komoltsev & Gulyaeva, 2022). I. Komoltsev, & N. Gulyaeva (2022). Brain Trauma, Glucocorticoids and Neuroinflammation: Dangerous Liaisons for the Hippocampus. *Biomedicines*, *10*, 1139. https://doi.org/10.3390/biomedicines10051139. Reproduced with permission from *Biomedicines* (Creative Commons Attribution 3.0 Licence).

Learning and memory are associated with the DH. The role of the HPC in memory was initially explored by James Papez in 1937 when he injected the rabies virus into the HPC of rodents and then saw its progression through the brain (Bhattacharyya, Gholipour et al., 2017). The HPC creates declarative, otherwise known as explicit memory (Tulving & Markowitsch, 1998; Eichenbaum, 2004). Declarative or explicit memory is information that can be readily recalled, e.g., knowledge of events and experiences in the external world. It helps form links between them (Eichenbaum,

2000). There are two types of explicit memory: episodic and semantic. While the former refers to first-hand experiences, the latter concerns facts and knowledge of the world (Stangor, 2014). CA1, CA3, and the DG are integral in acquiring contextual memories (Lee & Kesner, 2004). 'Declarative Theory' differentiates between the age of memories, believing that consolidated episodic memories are stored outside the HPC and in the neocortical areas. The study supports research on Henry Molaison (HM), who underwent surgery in 1953 to remove 2/3 of their hippocampi to elevate their extreme epilepsy convulsions (Scoville & Milner, 1957). This surgery made HM unable to convert short-term memories to long-term memories. Therefore, they started each day the same as the day before, as they suffered from anterograde amnesia. However, they discovered that some of Molaison's long-term memories were intact, confirming that long-term memories are stored outside the HPC.

Studies have shown that age-related improvements in episodic memory increase the hippocampal volume (DeMaster, Pathman et al., 2014). Riggins et al. examined the connection between hippocampal function and episodic memory in early years, precisely with four and six-year-olds (Riggins, Geng et al., 2016). The results showed similar functionality in both areas; however, three of the four regions within the HPC were positively associated with episodic memory in six-year-olds and negatively with younger children. In addition, three areas outside the HPC were positively associated with episodic memory in younger children and not with the older cohorts (Riggins, Geng et al., 2016). Therefore, it may be concluded that the HPC becomes integrated with other regions related to hippocampal memory in adulthood (Riggins, Geng et al., 2016).

The role of the DH has been long associated with spatial navigation. This was based on the initial discovery of 'place cells' by O'Keefe and Dostrovsky (O'Keefe & Dostrovsky, 1971). 'Place cells' have been recorded in rodents, monkeys, and humans during the past 36 years. It is believed that 'place cells' fire when a person is placed in or exploring a new environment. 'Place cells' present a sense of site of the location but not the orientation preference of the person (O'Keefe, Burgess et al., 1998). These 'place cells' are also impacted by the subject's motivation, as in the subject's movements, referred to as 'path integration'. These are linked to the EC, one of the prominent connections to the HPC. The discovery of these 'place cells' has

encouraged much research in spatial navigation in recent years (Moser, Moser et al., 1993; Moser, Moser et al., 1995; Holt & Maren, 1999; Pothuizen, Zhang et al., 2004; Lee, Lew et al., 2019).

This visuospatial memory (VSM) allows us to understand our environment and ourselves in relation to other objects, allowing us to navigate a particular space. An exciting study by Maguire et al. discovered that qualified taxi drivers had an enlarged posterior HPC. All London taxi drivers must pass a comprehensive exam on the map of London entitled 'The knowledge' to increase their familiarity with all routes in this large city. Compared with the hippocampi of non-taxi drivers, the hippocampi of taxi drivers showed a significant increase in grey matter (Woollett & Maguire, 2011). However, little is known about how hippocampal development impacts children's large-scale spatial navigation capabilities (Vieites, Nazareth et al., 2015).

1.7.2 Structural Differences in the Hippocampus in Those with Down Syndrome

Individuals with DS have a smaller HPC with fewer neurons (Weitzdoerfer, Fountoulakis et al., 2001). Post-mortem examinations of both foetuses and adults have shown reduced neuronal densities in many brain areas across a lifespan. The deficits in the HPC begin at 17–21 weeks with reduced neurons and an elevated level of apoptosis (Guidi, Bonasoni et al., 2008). Guidi et al.'s study also found that the brains of foetuses at 17–21 weeks of gestation show reduced cell proliferation in the DG (Guidi, Bonasoni et al., 2008). This is supported by Sylvester, who found that the DG in the brain of a DS foetus is not as developed as that found in the TD brain, creating a more significant gap between DG and the pyramidal cell layer (PCL) (Sylvester, 1983). There is also reduced myelination in the hilar region of the DG (Ábrahám, Vincze et al., 2012). The differences between the HPC of a TD foetus and that of a foetus with DS may be seen in Figure 1.17, where there are evidential differences between most parts of the HPC, including the DG, the ependymal surface (Ep), the inferior horn (IH), and the end folium (EFL).

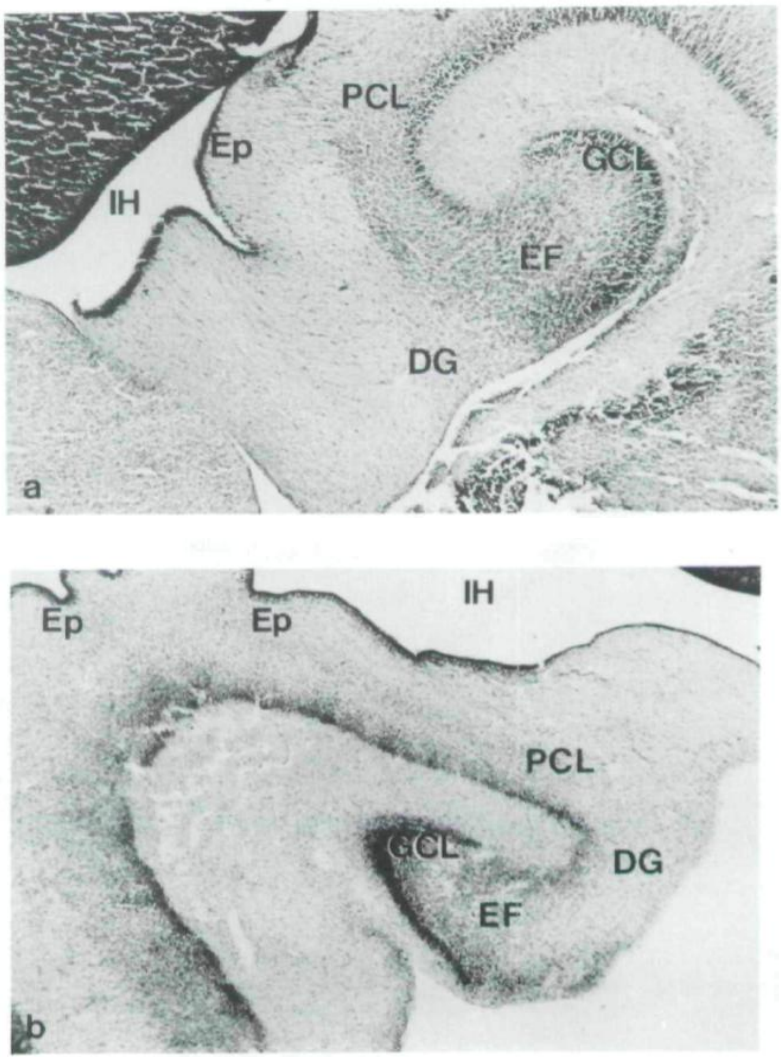


Figure 1.17. Coronal sections in the hippocampus of a TD foetus (a). The coronal sections in the hippocampus of a DS foetus (b). The DS hippocampus has fewer folds, while the dentate gyrus (DG) is small. There are differences in the ependymal surface (Ep), and the inferior horn (IH) is dilated. Sylvester also notes that the neuroblasts are less developed in the pyramidal cell layer (PCL) and the granular cell layer (GCL) (Sylvester, 1983). P. E. Sylvester (1983). THE HIPPOCAMPUS IN DOWN'S SYNDROME [https://doi.org/10.1111/j.1365-2788.1983.tb00294.x]. *Journal of Intellectual Disability Research*, *27*(3), 227–236. https://doi.org/10.1111/j.1365-2788.1983.tb00294. Reproduced with permission from John Wiley & Sons Books. (Blackwell Publishing Ltd.).

The intrinsic aberrations to the neuronal construction in the HPC and DG contribute to significant difficulties encountered by those with DS. These include altered synaptic plasticity and alterations in the structure of the dendrites. There is a significantly decreased density in the hippocampal dendritic spines in those with DS, which impacts the perceived learning capacity of those with the condition (Suetsugu & Mehraein, 1980; Takashima, Becker et al. 1981; Marin-Padilla & Marin-Padilla,

1982; Becker, Armstrong et al., 1986; Ferrer & Gullotta, 1990; Fiala, Spacek et al., 2002).

The physical structure of dendrites, their body, and their densities impact plasticity. A significantly reduced number of dendrites can lessen the potential sites for plasticity (Cramer & Galdzicki, 2012). Initially, dendritic expansion progresses in the first few months of life, regressing significantly in the second year in those with DS (Cramer & Galdzicki, 2012). This is supported by Weitzdoerfer, who agrees that a foetus with DS starts with an average neuronal density but has impaired dendritic spines with few synapses (Weitzdoerfer, Fountoulakis et al., 2001). Synaptogenesis is a vital component in connecting networks in brain development. Therefore, alternations in the HPC will impact maturity (Hanson, Blank et al., 2007). A decrease in the formation of synapses onto CA3 neurons indicates that those with DS have challenges with pattern completion, pattern separation and contextual pattern separation (Lee & Kesner, 2004; Vazdarjanova & Guzowski, 2004). Pattern completion, which takes place in the CA3 region of the HPC along with independent memories, allows the reactivation of cells to complete a pattern (Rolls, 2013). Pattern separation occurs in the DG and is the process that reduces the overlapping of neuronal activity that corresponds to similar experiences (Rolls, 2013; Madar, Ewell et al., 2019). It is part of episodic memories and is responsible for forming distinct entities from similar information (Yagi, Lee et al., 2022). Contextual pattern separation is the ability to separate distinct entities from similar environmental situations (Yagi, Lee et al., 2022). This leads to limitations in memory capacity in people with DS (Hanson, Blank et al., 2007).

Substantial evidence has been found in mouse models to show that plasticity is severely affected in mice with DS (Cramer & Galdzicki, 2012). The Ts65Dn mouse model has been instrumental in demonstrating the many phenotypes found in humans with DS. Mouse models have been used to help understand this syndrome's physiological and behavioural characteristics. The most significant advancement was the mouse model Ts65Dn, developed in 1993 (Davisson, Schmidt et al., 1993). It is not a replica; however, the mice share many physical and behavioural characteristics of those with DS (Tyler & Haydar, 2013). The "Ts65Dn broke open the door," said Tarik Haydar from Boston University. The Ts65Dn mouse model displays enlarged

boutons and dendritic spine heads in cortical and hippocampal neurons (Zigmond, Rowland et al., 2015). The mouse chromosome 16 is trisomic and reflects many of the genes exhibited from chromosome 21, including the difficulties encountered and associated with hippocampal dysfunction (Reeves, Irving et al., 1995; Demas, Nelson et al., 1996; Hyde & Crnic, 2001). For example, significant reductions were found in the synaptic function of the excitatory connections and inhibitory inputs to the pyramidal neurons in the CA3 of the HPC in Ts65Dn animals (Hanson, Blank et al., 2007). This is believed to contribute to memory deficits in those with DS.

1.7.3 Alterations in Hippocampal Function in Those with Down Syndrome

The Ts65Dn mouse model shows reduced neurons and synapses in the DG, CA1, and CA3. There is reduced connectivity in the CA3 and a reduced perforant pathway, the Schaffer collateral, LTP, and disruption to excitatory and inhibitory signalling (Kurt, Kafa et al., 2004; Best, Cramer et al., 2012; Deidda, Parrini et al., 2015). Deficits accompany these abnormalities on rodent hippocampal tasks requiring spatial navigation or context discrimination (Reeves, Irving et al., 1995; Hyde & Crnic, 2001; Sérégaza, Roubertoux et al., 2006). Hippocampal studies have typically used the Ts65Dn mouse model to depict the challenges in learning and memory (Reeves, Irving et al., 1995; Demas, Nelson et al., 1996; Sago, Carlson et al., 2000; Hyde & Crnic, 2001). Although there is limited insight into the biological basis for cognitive failure, explicit memory abnormalities indicate the HPC's involvement (Carlesimo, Marotta et al., 1997; Caselli & Tonucci, 2000; Pennington, Moon et al., 2003). Explicit memory in Ts65Dn mice exhibit several features characteristic of DS, including impairment of hippocampal function as evidenced by impaired spatial memory (Reeves, Irving et al., 1995; Demas, Nelson et al., 1996; Holtzman, Santucci et al., 1996; Sago, Carlson et al., 2000) and context discrimination (Hyde & Crnic, 2001). Synaptic connections are excitatory and formed at the heads of the dendritic spines; therefore, the structure of these spines and their shape are significant. Several studies have confirmed that reduction in excitatory synapses in those with DS has led to the abnormal structure of synapses in the HPC (Belichenko, Masliah et al., 2004; Kurt, Kafa et al., 2004; Hanson, Blank et al., 2007).

Additional studies based on these mouse models have shown deficient performance in visuospatial tasks, such as the Ts65Dn in the Morris water maze, which depends on the HPC (Reeves, Irving et al., 1995; Edgin, Mason et al., 2010). The maze consists of a container filled with water and a submerged, hidden platform (Morris, 1981). The mouse must learn where the platform is and memorise its location to escape the water. However, when the HPC is lesioned, it interferes with their ability to find the platform. Such tests have led to more detailed studies on the neuropsychology and the HPC of children with DS (Reeves, Irving et al., 1995; Edgin, Mason et al., 2010).

There are also considerable challenges in contextual pattern separation in DS mouse models (Clark, Fernandez et al., 2017). Such models have shown changes in dendritic spine construction in the HPC, less neurogenesis and LTP in the DG, and excitatory connectivity in the DG-CA3 (Kleschevnikov, Belichenko et al., 2012; Witton, Padmashri et al., 2015; Clark, Fernandez et al., 2017). Granular cells are also crucial for pattern separation (Leutgeb, Leutgeb et al., 2007), and there can often be disruption to these in those with DS. In 2014, Smith, Kesner and Korenberg tested object, spatial, and unusual configurations in Ts65Dn/TnJ mice (Smith, Kesner et al., 2014). The Ts65Dn mice showed impairment in recognising objects in contexts with many spatial and contextual cues (Clark, Fernandez et al., 2017). This also interfered with the representations of familiar objects. However, the mice were unaffected when short-term object recognition did not incorporate contextual cues (Clark et al., 2017). This suggests that other areas in the medial temporal regions of the brain, such as the perirhinal cortex, are intact in children with DS.

Several studies show that DS groups of children have difficulties with tasks sensitive to hippocampal disruption relative to mentally aged-matched (MA-matched) controls (Pennington, Moon et al., 2003; Edgin, Mason et al., 2010; Ribordy, Jabès et al., 2013; Costa, Purser et al., 2015). There has also been a substantial examination of LTM deficits in those with DS (Ellis, Woodley-Zanthos et al., 1989; Caltagirone, Nocentini et al., 1990; Devenny, Hill et al., 1992). Ellis et al. used picture books to examine non-verbal LTM. The group with DS could not recognise pictures and their locations, indicating alterations in hippocampal function (Ellis, Woodley-Zanthos et al., 1989). Prevalent deficiencies characterise DS in episodic memory and

learning that can have widespread adverse implications for daily functioning (Carlesimo, Marotta et al., 1997; Edgin, Pennington et al., 2010; Spanò, Peterson et al., 2015; Clark, Fernandez et al., 2017). Carlesimo et al. examined implicit and explicit memory and found altered functions in episodic memory, which are integral in creating LTM specific to each person, event, and experience (Carlesimo, Marotta et al., 1997).

Mangan has previously studied such altered functions in the HPC of individuals with DS, focusing on the learning and recall of children aged 16–30 months (Mangan, 1992). This involved a DS cohort and chronologically age-matched (CA-matched) control group that engaged with three spatial tasks, one of which was place learning and recall. The DS group found this task more challenging than their CA-matched peers on the learning component of each task; however, they experienced increased difficulty in the recall aspect.

People with hippocampal damage have impaired abilities in relational mapping, that is, connecting objects to specific spatial environments over shorter intervals (Clark, Fernandez et al., 2017). In a recent study, Clark and colleagues developed a set of novel tasks measuring medial temporal functions, including visual, spatial, and temporal memory (Clark, Fernandez et al., 2017). Seventy-six participants were involved in the study, divided between an MA-matched cohort (n=48) and a DS group of children and young adults (n=28). The DS group was unimpaired on short-term and configural recall tasks with no spatial or temporal element (Clark, Fernandez et al., 2017). However, the DS cohort struggled when the objects relied on contextual cues and when objects were expected to be recalled in order (Clark, Fernandez et al., 2017). The findings are consistent with the Ts65Dn mouse model studies. Short-term spatial memory may correspond with general cognitive ability and a particular strength in those with DS (Edgin, Pennington et al., 2010; Yang, Conners et al., 2014). However, there are significant challenges in spatial and temporal contextual pattern separation skills mediated by the HPC. CA1 completes the transsynaptic circuit, indicating that this region is essential in temporal order judgment (TOJ) (Kesner, Morris et al., 2012).

The HPC binds memories to spatial and temporal contexts (Diana, Yonelinas et al., 2007). The HPC has traditionally been associated with LTM, but as described

above, it has also been associated with relational connections, objects in contexts, and short-term associations (Bontempi, Laurent-Demir et al., 1999; McGaugh, 2000; Hammond, Tull et al., 2004; Bontempi & Frankland, 2010). Following the conclusions made by Smith et al. and Clark et al., those with DS have difficulty with contextual-based visual, spatial, and temporal tasks (Smith, Kesner et al., 2014; Clark, Fernandez et al., 2017). Evidence suggests that some medial temporal regions, specifically the perirhinal cortex, are intact. Pennington similarly confirms hippocampal atypical functionality as a common characteristic in DS patients (Pennington, Moon et al., 2003). However, they also emphasise the need for studies to identify when the altered function begins in order to find an appropriate means of intervention.

1.8 Interconnective Altered Functions in Individuals with Down Syndrome

As described, people with DS suffer from impairments in planning, memory, and decision-making (Pennington, Moon et al., 2003; Lanfranchi, Jerman et al., 2010; Lavenex & Banta Lavenex, 2013; Grieco, Pulsifer et al., 2015; Clark, Fernandez et al., 2017). The main reason for this is due to the altered functions of the HPC and PFC (Nadel, 2003; Pennington, Moon et al., 2003; Nelson, Leibenluft et al., 2005; Lott & Dierssen, 2010; Anderson, Nielsen et al., 2013; Ruiz-Mejias et al., 2016). It has been found that while dialogue between the two areas is essential, higher connectivity is related to low IQ. Neuroimaging studies have confirmed this atypical connectivity in individuals with DS. In a study by Maria Gonzaleza et al., the Ts65Dn mice showed a hyper-synchronised neural activity between the PFC and the HPC. They tested several states, including wakefulness, sleep, and performance on memory tasks (Alemany-González, Gener et al., 2020). The mice displayed high levels of communication during memory acquisition, familiarisation, and retrieval (Alemany-González, Gener et al., 2020). However, the mice showed deficits in declarative memory, particularly spatial navigation. A further study was undertaken by Chang et al. to explore the circuitry between the medial PFC and HPC. The team aimed to show ‘altered neural dynamics’ in the PFC-HPC of mouse models Dp1Tyb, Dp10Yey, and Dp17Yey, which cover all homology regions analogous to people with DS (Chang, Bush et al., 2020). The Dp10Yey mouse model showed deficits in spatial working memory (SWM), while

the Dp17Yey model had difficulties in decision-making overall, it showed normal cognitive function similar to the wild type (Chang, Bush et al., 2020).

A recent study by Shi-Yand Xu et al. reported altered connectivity between the motor cortex and the PFC (Xu, Lu et al., 2020). Seven children with DS were recruited, gender and MA-matched with 6 TD children. Within this study, they played a computer game called 'Catch the Hamster', where four hamsters and their burrows were presented. The child had to show which hamster came from which hole at each task. The child had 32 trials over 20 minutes. The trials were carried out using functional neon-infrared spectroscopy (fNIRS) software. Thirteen channels were recorded from the motor cortex to the PFC. Children with DS performed at a lower function than their TD counterparts.

The Arizona Cognitive Test Battery (ACTB) is one of the main tests exploring the link between neuronal and dendritic structures and synaptic functions. The ACTB was designed to assess the cognitive functions of those with DS by evaluating the HPC, the PFC, and the cerebellar regions (Edgin, Mason et al., 2010). The ACTB has been designed to be a non-verbal task, apply to various ages, have specific relationships to brain function, and test several skills (Edgin, Mason et al., 2010). The tasks involved in this test include picture pointing, Corsi-like tests that include recreating a particular order, spatial pattern completion tasks, verbal skills, and scales of independent behaviour. The design of this test and the results found in such studies and others show the extremes in cognitive ability across this cohort (Edgin, Mason et al., 2010).

The interconnectivity between the cerebellum, PFC, and HPC has been extensively documented, while the atypical structures of these brain regions in those with DS have also been detailed. The role of these regions in visuospatial working memory (VSWM) has also been noted. The following review, details some of the most prominent research in this area with children and adults with DS.

1.9 Visuospatial Working Memory in Those with Down Syndrome

Visuospatial ability is a broad term that depicts how we perceive and process visual information within the environment, combining this information with other senses and experiences to attain meaning (Lohman, Pellegrino et al., 1987; Yang, Conners et

al., 2014). Visuospatial working memory (VSWM) can retain these visuospatial images for a short period (Rizzo & Vecera, 2002). The VSWM, for example, allows us to recognise shapes and colours, their location, and their movement. While functional differences have been previously noted in the brain areas responsible for VSWM (the PFC, HPC, and cerebellum), several studies have reported that VSWM is a relative strength in those with DS. However, it is essential to remember that there are still functional differences compared to MA-matched TD groups (Bird Elizabeth & Chapman Robin, 1994; Costa, Purser et al., 2015). Visuospatial working memory is often considered a strength in this cohort because it is compared with verbal short-term memory, which presents more challenges in people with DS. Therefore, VSWM-making abilities are the most remarkable.

In a recent editorial for *Brain Sciences*, Lanfranchi et al. stated that there are patterns of strengths and weaknesses in the visuospatial skills of those with DS (Lanfranchi, Meneghetti et al., 2022). They similarly recognised that visuospatial abilities in those with DS have 'peaks and troughs' (Lanfranchi, Meneghetti et al., 2022). For example, spatial sequencing memory and visuospatial integration are less impaired than rotation skills and visual and spatial simultaneous memory. Such disparity in the visuospatial skills of those with DS was also found in a comprehensive review by Yang et al. (Yang, Conners et al., 2014). In this review, Yang and colleagues included 49 studies from 1987 to 2013 to explore the strength of these visuospatial skills. Yang et al. considered only studies with MA-matched DS children with TD children, and those studies with CA-matched groups were excluded, as were studies that involved young children aged 1–3 years.

Several research projects exposed fundamental issues in carrying out studies with this cohort. Research projects with DS children and adults are restricted by numbers, with few committing to partake in these studies. Consequently, there is often an extensive age range with a broad spectrum of abilities involved in the studies. The underlying difficulty is that there is no one developmental trajectory for those with DS. Therefore, several contrasting methodologies have been used in numerous studies to assess these children at baseline, with some researchers using multiple methods to MA-match the cohorts.

To strategically access these studies, Yang et al. reviewed ten spatial factors put forward by Lohman et al. in 1987. These ten abilities included visualisation, spatial orientation, the flexibility of closure, closure of speed, spatial relations, spatial scanning, perceptual speed, serial integration, visual memory, and kinaesthetic (Lohman, Pellegrino et al., 1987). Yang et al. also considered the five spatial factors mentioned by Carroll in *Human Cognitive Abilities*. In contrast, Carroll identified just five abilities: visualisation, spatial relations, closure flexibility, closure speed, and perceptual speed, with spatial memory considered separately in this approach. Yang et al. concluded that visuospatial memory, visuospatial construction, closure, mental rotation, and wayfaring categorisations would best apply to the studies examining the visuospatial abilities of those with DS. Visuospatial memory refers to how we can attain information on an object and its spatial location (Yang, Conners et al., 2014). Yang et al.'s visuospatial construction is like that of Lohman et al. and Caroll's idea of visualisation. It is the ability to construct an object even if only the parts are given. Mental rotation is the ability to rotate 2D and 3D objects, while closure is building a whole thing out of components and deconstructing them into individual pieces. Finally, wayfaring was included in this review, as it is often a feature incorporated in such studies. Wayfaring refers to finding and following a route from beginning to end (Yang, Conners et al., 2014). Visuospatial memory was examined in many of these research projects, and therefore, Yang et al. subdivided visuospatial memory into four subsections. As shown in Figure 1.18, these include spatial sequencing memory, spatial simultaneous memory, memory for locations, and spatial working memory. Each of these will be discussed in the sections that follow.

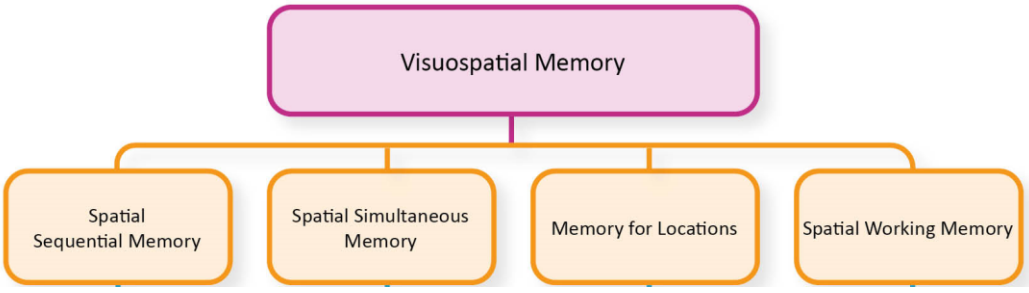


Figure 1.18. A visual representation created from the subdivision of visuospatial memory, as seen in (Yang, Conners et al. 2014).

1.9.1 Spatial Sequential Memory

Spatial, sequential memory (SSM) is the order in which spatial information is presented (Yang, Conners et al., 2014). In many of the articles examined by the reviewers, the Corsi Blocks test was the most widely used with those with DS, both with physical Blocks and computerised versions (Milner, 1971). The facilitator presents a sequence of blocks to the participant, tapping a pattern on the blocks, which the participant then replicates. The built-in complexity depends on the number of trials and blocks used. Some computerised derivatives of the test have been devised to include animation, encouraging the children to concentrate and engage more with the task (Lanfranchi, Cornoldi et al., 2004). The Corsi Blocks test is believed to yield the most consistent results, according to Yang et al. (Yang, Conners et al., 2014). Out of the 17 studies making 24 comparisons between MA-matched DS and TS groups, it was perceived that SSM corresponds to the overall cognitive ability of both groups (Yang, Conners et al., 2014).

Interestingly, 20 of the 24 comparisons showed no significant difference between the groups. These studies used several contrasting measures to MA-match the cohorts, including general cognitive ability, receptive vocabulary, and reasoning skills. Several derivatives/variants of the Corsi Block tests were used, and the MA-matched participants ranged from 4 to 7 years, which created difficulty in comparing the studies. However, it was identified in three studies that the DS cohort experienced more challenges in SSM than the TD group, perhaps challenging this aspect of visuospatial strength (Vicari, Bates et al., 2004; Vicari, Bellucci et al., 2006). Each was MA-matched according to general cognitive ability (MA from WISC-III, Stanford Binet, Bozzo, & Mansueto Zecca, 1993; Wechsler, 2002; WAIS-III, Wechsler, 2004). In one study, Duartes et al. used two MA-matched strategies, one general cognitive ability and the other receptive vocabulary (from WISC-II and WISC-III, and matched receptive language from Peabody Picture Vocabulary Test, PPVT) (Dunn, 1997). While being MA-matched on general cognition revealed that the DS group experienced more difficulty, there was no difference when they were MA-matched on receptive vocabulary. Remarkably, only one study by Laws et al. found DS participants to perform better than the TD cohort, who were MA-matched through

receptive language (Laws, 2002). While only one study showed that the DS cohort performed better, SSM is still considered a strength in relation to other cognitive abilities.

A study by Frenkel and Bourdin in 2009 could account for differences between study results. They discovered that both groups performed at the same level when MA-matched at three years old. However, when the MA-matched increased to 8.5 years, the TD performed more successfully than the DS cohort on the Corsi Blocks test (Frenkel & Bourdin, 2009). Therefore, indicating that those with DS had an atypical developmental trajectory in SSM.

A more recent study by Meneghetti et al. examined the role of spatial sequence in path learning of those with DS (Meneghetti, Toffalini et al., 2021). Path learning affects everyday life and the ability to find one's way around. Research has shown that those with DS can learn and retrieve spatial information from an egocentric (person) point of view when shown the path either in person or on a video (Meneghetti, Toffalini et al., 2021). The aim of this study was two-fold: firstly, to assess the groups' performances on the paths under contrasting conditions by replicating the pathway by looking at someone doing the path and, secondly, when instructed orally (Meneghetti, Toffalini et al., 2021). The additional aim was to assess if this reflects the participant's cognitive abilities. There were 62 involved in the study, 30 with DS and 32 TD participants. They were matched using the PPVT on receptive vocabulary (Dunn, 1997; Italian adaptation by Stella et al., 2000; PPVT-R). This involved the examiner referring to one of four pictures and the participants pointing to which picture they thought it was. Secondly, Raven's Colored Progressive Matrices were also used (Raven, Schwarzwald et al., 1998); Italian adaptation by Belacchi et al., 2008; CPM). The participants selected one part from six pieces they thought were missing from a matrix.

The study included 36 trials with increasing difficulty in testing visuospatial elements. For the actual research, a 4x4 matrix was created on the floor with 16 grids that formed the basis of the pathways. Ten trials were conducted for each learning condition. In both groups, the observation learning was more robust. Interestingly, both groups encountered difficulties with the oral instruction. This study confirms

that those with DS can learn a long sequence of steps following spatial sequencing capabilities and that verbal instruction is less effective.

1.9.2 Spatial Simultaneous Memory and Memory for Locations

Spatial simultaneous memory has also been explored extensively in relation to those with DS. An example of a task exploring such is when a participant is shown squares filled with a specific colour while others are left blank or filled with a contrasting colour. Before the colours disappear, the participant can view the pattern for a prescribed time. They are then invited to identify what squares had the target colour in any order (Yang, Conners et al., 2014). Out of seven studies, it was confirmed that pattern recall was not a strength in the DS cohort, regardless of whether consistent MA-matched methods were used. Two studies, one by Lanfranchi et al. and another by Hick, Botting, and Conti-Ramsden, found that the DS and TD cohorts performed at a similar level (Lanfranchi, Cornoldi et al., 2004; Hick, Botting et al., 2005). In an additional study by Carretti et al., three groups participated, two of which were matched on receptive vocabulary and the other on general cognitive ability. The result showed that the DS cohort found the task more challenging than the TD groups (Frenkel & Bourdin, 2009; Lanfranchi, Carretti et al., 2009). Like results found in SSM, Lanfranchi and Carretti et al. stated that pattern recall might also develop at a different rate in the DS cohort than in their TD counterparts (Lanfranchi, Carretti et al., 2009).

This was explored further in a longitudinal study by Hick et al. in 2005. The children were presented with sharks occupying half of the cells. When they were first MA-matched through a non-verbal test at 4.3 to 4.5 years, there was no difference in the two groups' abilities at pattern recall; however, the DS group showed a disadvantage when tested six months and 12 months later. Another test by Frenkel and Bordin included a pattern recall test (Frenkel & Bourdin, 2009). Their sequence representation appeared again with one unfilled square, and the participants pointed at the empty square. In additional tests, the participants had to process the configuration in a minimal amount of time and therefore reduced memory demands, as they pointed out. The evidence here is inconclusive, with those with DS being inconsistent or lower than developmental levels (Yang, Conners et al., 2014).

Memory for locations has been an additional area of research used to examine the visuospatial capabilities of those with DS. This involves participants being presented with different items and recalling their location sometime later (Yang, Conners et al., 2014). These differ from the pattern recall tests with no time-lapse, and the areas are filled automatically. When familiar pictures and objects were used, both DS and TD groups performed comparatively (Zucco, Tessari et al., 1995; Vicari, Bellucci et al., 2005). However, when abstract words and nonsense pictures were used, the DS cohort found the task more complex than the TD group.

1.9.3 Spatial Working Memory

Spatial working memory (SWM) involves taking in information, processing it, and manipulating it to apply to learning. Eighteen comparisons were made between ten studies in this section by Yang et al. In this task, the Corsi Blocks test was most frequently used; however, the participants had to put the sequence in reverse. Therefore, this requires the storage of the sequence and spatial location plus processing the series by reversing its presentation. Vicari et al. found that those with DS encountered more challenges with the task than the TD group even when MA-matched on non-verbal skills. Lanfranchi et al. used a derivative of this test with frogs jumping to appeal to younger children, and they found no difference between both groups. Perhaps this encouraged the children to engage more with the task and heightened their attention (Lanfranchi, Cornoldi et al., 2004).

Other studies used the Cambridge Neuropsychological Test Battery (CANTAB) spatial working memory test (Fray, Robbins et al., 1996). The participants are presented with 3–8 boxes to locate a token. Once they touch the box and open it, it closes again. The participant must remember which boxes they have visited, as the tokens will appear in different boxes each time. Pennington et al. (2003) found no difference between the MA-matched groups. However, Visu-Petra, Benga, Tincas, and Miclea found that those with DS had fewer correct answers than the TD cohort (Visu-Petra, Benga et al., 2007).

Lanfranchi et al. (2004) used a derivative of the Corsi Blocks Backward test, called the backward pathway task, and labelled it a medium-low control task (Lanfranchi, Cornoldi et al., 2004). In addition, the starting position task involved

participants being shown one or two frogs going through a 4x4 chessboard. They are then asked to remember the frog's starting position –the medium-high task. The higher control test required the participants to remember the frog's starting point and tap the table every time the frog jumped into a red square. They found that TD and DS children performed similarly on the medium-low and backwards pathway tasks. However, differences began to emerge on the highest tasks.

Lanfranchi believes that the more control a WM task requires, the more likely it is for those with DS to encounter challenges, i.e., when processing demands are high (Yang, Conners et al., 2014). A summary of the tests identified by Yang et al. may be seen in Figure 1.19.

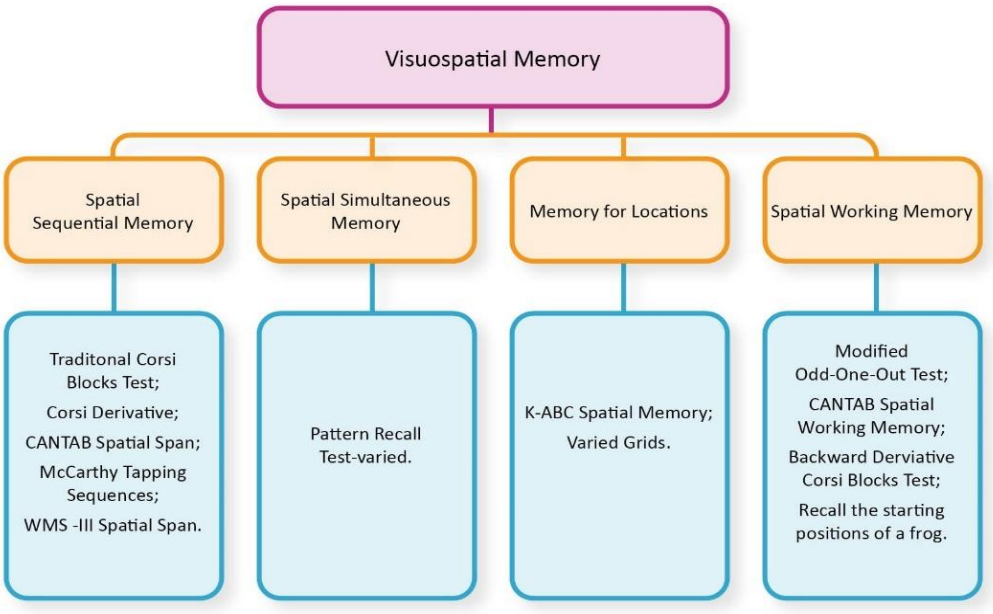


Figure 1.19. A figure created for this thesis from the visuospatial memory components, and the tests most associated with them, as identified by Yang et al., is subdivided further. (Yang, Conners et al., 2014).

1.9.4 Visuospatial Skills in Those with DS in Summary

The above studies show SSM to be a strength in those with DS (Meneghetti, 2002) and reflective of general knowledge. However, simultaneous spatial memory is either the same or more challenging, with SWM being a particular difficulty. A recent study by Carretti et al., published in 2021, explored the developmental trajectory involved in SSM and simultaneous spatial memory. This study aimed to examine the development of these skills from a chronological and developmental perspective. As

predicted, the performance on spatial-simultaneous WM tasks was more challenging for both TD and DS groups, with the DS group showing a developmental progression until the age of 13 with limited progression from that point (Carretti, Meneghetti et al., 2022). The developmental stop in the early teenage years confirms the expected cognitive decline from this age (Dykens, 2000). However, many studies above illustrate too much variety in the testing methods used, even within each visuospatial category. In addition, there is a diversity in how the cohorts are MA-matched. The review conclusions indicate that, as Lanfranchi identified at the start of this section, there are troughs and flows in the visuospatial skills of those with DS. They have performed at the same level or have encountered more challenges than their TD counterparts. Measuring spatial sequencing abilities with Corsi Blocks brings a low processing load, which is perhaps why this is the most widely used task with this cohort. Consistency with MA matching needs to be addressed; there is a need to test more significant numbers while also more longitudinal studies. Yang concluded that if correct intervention strategies were found, it could significantly impact the quality of life for this cohort.

1.9.5 Developing Visuospatial Skills in Those with Down Syndrome: Intervention Studies

Since Yang's review, several studies have focused on advancing memory skills in children with DS by improving visuospatial skills through rehearsal. One study by Bennett and colleagues focused explicitly on VSM. The study included participants taking computer-based training to improve visuospatial STM (Bennett, Holmes et al., 2013). This involved 21 children aged 7 with DS being assigned to the activity or randomly to the control group. It was found that after a 12-week training period, prepared and unseen visuospatial tasks improved considerably and remained after four months. As pointed out by Costa, Bennett's study is the only one to examine the implication of WM training on both STM and WM of visuospatial elements (Bennett, Holmes et al., 2013; Costa, Purser et al., 2015). Within the proposed intervention in this current study, a similar online music programme was used to assess its impact on the VSM of children with DS.

Costa et al.'s study investigated the effect of school-based learning on STM and WM of two participants with DS (Costa, Purser et al., 2015). Their research included seven computerised STM and WM games: four involved the storage of visual information, two involved manipulations and storage of visual information, and one included both (Costa, Purser et al., 2015). This research expected a positive outcome on both the STM and WM in visuospatial skills and transferring these skills to VM skills. The participants used eight paper and pencil-designed training tasks in this study to enhance their VSWM skills. Over six weeks, they played two games at each of the 12 sessions for 40 minutes (Costa, Purser et al., 2015). The training was an adaptation of Passolunghi and Costa's version in 2014. There were four VSWM games and four visuospatial STM games.

For the assessments, Costa et al. used the pathway recall method developed by Lanfranchi to test visuospatial STM (Lanfranchi, Cornoldi et al., 2004). The child was shown the frog's pathway on a 3x3 or 4x4 grid and had to replicate it. There were eight trials and four difficulty levels, with two attempts on the more difficult levels. Lanfranchi et al.'s pathway backward recall was used to test VSWM. This was like the previous test; however, the child had to remember the reverse order pathway (Lanfranchi, Cornoldi et al., 2004). There were, again, four levels of difficulty. Costa et al. also included a selective pathways task, again based on that of Lanfranchi et al. 2004. The participants were shown two routes of the toy frog on a 4x4 grid, and they replicated the starting point of the frog. There were four levels, levels 1 and 2, with two and three steps, and at levels three and four, there were three and four steps. This task included a visuospatial dual task adapted from Lanfranchi (Lanfranchi, Cornoldi et al., 2004). Within this task, the child had to remember the frog's starting position on a 4x4 grid wherein one of the 16 cells was red; when the frog jumped to the red square, the child had to tap on the table.

Costa also included a VSTM task and a VWM task. These were again based on Lanfranchi's tasks, including the forward word recall, which required a list of 2–5 words the child must recall. The child repeated each list in reverse order to test verbal WM in the backward word recall. The test was coupled with a selective recall test where the child repeated each list's first word after the entire series was presented. A verbal dual-task was also inspired by Lanfranchi, where the child was given 2–5

syllable words and then asked to repeat the first word on the list and then tap the table when the word 'ball' was presented (Lanfranchi, Cornoldi et al., 2004).

Both participants in the study saw a marked improvement in visuospatial scores after the training process and the results were better in areas where they were initially significantly impaired. The study was limited due to having just two participants involved. Therefore, more children must be tested in future studies. In addition, the changes were only tested immediately after and not a few months later to see if there were lasting effects. However, there is evidence that the visuospatial training impacted verbal abilities more than the other skills. The study also shows the variance in capabilities between those with DS. Costa et al. summarised one of the most exciting aspects of this study: that “WM abilities can be improved, and visuospatial training can be effective for children with DS" (Costa, Purser et al., 2015).

Another study by Lanfranchi et al. also examined the potential of administering training to improve the spatial-simultaneous aspect of WM (Lanfranchi, Pulina et al., 2017). The effects of the training were specific to spatial-simultaneous working memory. However, it was hoped this intervention would develop near and far transfer of skills that could impact related visuospatial domains such as spatial sequencing and visuospatial abilities. Parents trained one group, while a psychologist managed the other group. Thirty-nine children and adolescents with DS were divided between the two groups. The spatial simultaneous working memory tasks that were issued included the following. The participants were given eight seconds to process two or three green-coloured squares on a 2x2, 3x3, or 4x4 matrix. They were then immediately asked to recall the positions on a blank matrix. There were varying levels of difficulty depending on the size of the matrix.

For the active task, the participants were then shown a matrix of contrasting sizes containing two or three red squares; some boards also had a blue square. The participants were then asked to remember the positions of the red squares pointing to their locations. Additionally, they had to tap on the table when a blue square appeared. A test was used to assess the near transfer of skills from the training of SSM. The participants were to replicate a path taken by a frog on various matrixes immediately after seeing it. There were four levels of difficulty. The near transfer of skills examined general visuospatial abilities using geometric puzzles (NEPSY battery;

Kemp, 2011) and a standard memory questionnaire. The participant was given a large grid containing six shapes, and an additional two shapes outside the grid. The children were asked to match the two shapes outside the grid with two shapes inside the grid. There was a series of 12 trials; each trial lasted 45 seconds, and the participant was awarded full marks if both shapes matched and just one if they matched one shape.

The questionnaire comprised 16 questions, where the parent was asked to comment on everyday activities. For example, 'Can your child remember short songs or rhythms? (Cornoldi, Venneri et al., 2003). This was then assessed on a Likert-type scale where one was never or almost never, and four was always or almost always.

There were improvements in both groups in the spatial simultaneous WM tasks. However, the time at which they showed the impact of the intervention differed. While the 'expert' group showed immediate improvement, the parent-led intervention showed improvement in the post-follow-up period. The gradual improvement could be due to the difference in the interaction between the parent and child following the intervention programme. Interestingly, both groups performed the same in the active spatial simultaneous task; both performed better post-intervention. There were also improvements to both near and far transfer skills.

There was some improvement in WM regarding the near transfer effects, which was led by the spatial sequencing task. They performed better in the post-test than in the pre-test, which they maintained in the follow-up. The geometric puzzle results improved from the pre- to post-test and at the one-month follow-up. These are significant findings, as they show that parents could effectively deliver the intervention at home. The standard questionnaires resulted in contrasting findings. These improved from pre-follow-up in the expert group, which showed parents the benefits of their children taking part in the training. However, in the parent-led group, there was a dramatic dis-improvement. This likely resulted in the parent becoming increasingly aware of their child's limitations that may not have been emphasised before the intervention.

An additional study by Herrero et al. explored autonomous technologies to enhance attention, visuospatial STM and visuospatial processing in those with DS. Over three months, the mobile applications were used with 26 children with DS and TD children. The apps included Bubbles, which tests selective attention, 'Pair and

Learn,' which works on visuospatial STM; and Tangram, which works on visuospatial processing, all of which were developed by Smile and Learn (Herrero, Theirs et al., 2018)

The participants involved in the study were requested to use the applications one hour a week, with one 20-minute session awarded to each of the applications over three months. Only 14 out of the 26 participants complied with all aspects of the intervention. Therefore, the researchers focussed on the results of those who were fully compliant. There were significant improvements in performance on the Pairs and Learn and the Tangram; however, there was a slight improvement on the Bubbles application. The lack of progress in the Bubbles task could be due to the quick response demand. This has been proven previously to be a difficulty for those with DS (Devenny, Hill et al., 1992; D'Souza, Booth et al., 2016; Herrero, Theirs et al., 2018). The conclusions of this study show that engaging with an intervention, in this case, technology could enhance specific cognitive skills in this cohort. The research found relational evidence between the apps Bubbles and Pairs, and Tangram; however, not Bubbles and Tangram. The lack of relationship between the latter two could be because of Tangram's lack of time constraints, even though it did need attention. The relationship between Pairs and Learn and Bubbles could be that selective attention could lessen the cognitive load and enhance the short term visual VSM. The final relationship to be examined was Pairs and Learn with Tangram, which is believed to use comparable cognitive functions and similarly where visuospatial short-term memory can guide behaviour. On reflection, this study emphasises the potential importance of technological or alternative forms of intervention and their implication on the cognitive development of individuals with DS.

In examining the temporal and medial memory system, Clark et al. have identified the potential of intervention studies and their impact on the trisynaptic loop to enhance the cognitive skills of individuals with DS (Clark, Fernandez et al., 2017). To understand the workings of this cohort's spatial and temporal skills, the researchers developed novel tasks to access the medial temporal functions. In rodent literature, it is believed that the CA1 completes the trisynaptic loop and is considered an essential region in organising events depending on their occurrence. It has been suggested that the HPC, as well as having a significant role in LTM, has a role in rapid

multi-modal associations (Watson, Voss et al., 2013; Clark, Fernandez et al., 2017). In this study, the researchers explored this overlap between temporal and visuospatial functions in the HPC.

The novel tasks included a parade of well-known objects, as well as placing toy jacks in set sequences on a board with altered contexts, using different coloured matts and lures. The DS group performed well on the spatial recall tasks that involved recognising the spatial positioning of toy jacks when contextual cues were absent. However, they encountered difficulties with memory that relied on spatial and temporal contexts (Clark, Fernandez et al., 2017). This study confirms that those with DS face similar difficulties as those with lesions on the medial temporal lobe, showing that their STM for individual objects is intact (Clark, Fernandez et al., 2017). Therefore, temporal order may be one such challenge in this cohort. This study also points to the potential exploration of the trisynpatic loop, which may reveal further the implications of such atypicality in other cohorts. It is suggested that this should be the focus of other intervention strategies. Atypicalities are also evident in the neurochemistry of people with DS, which will form the focus of the following sections.

1.10 Altered Neurochemistry in the Down Syndrome Brain

Cortisol targets the limbic brain as it is linked with emotions and rewards (de Kloet et al., 2009). Cortisol is a glucocorticoid hormone produced by the adrenal glands. While it affects all the organs in the body, it has often been called the ‘stress hormone’, as it is linked to stress. In response to stress, CRH is released from the hypothalamus, which, in turn, leads to the release of ACTH from the pituitary gland (Kaimal, Ray et al., 2016). This stimulates cortisol release from the cortex of the adrenal gland into the bloodstream. Its functions in the body include supporting vascular responsiveness and maintaining glucose production from protein (Västfjäll, Juslin et al., 2012). Cortisone is considered to be linked to stress which can cause the natural, flight-fright-freeze response to stressful situations. Encountering stressful situations, therefore, increases cortisol levels. While short-lived, acute stress is necessary to protect us from potentially harmful situations by maintaining a steady energy supply and keeping the body and brain on high alert. Prolonged or consistently elevated cortisol levels can harm some biological systems, such as the cardiovascular,

gastrointestinal, and immune systems and lead to ill health, including anxiety and depression (Västfjäll, Juslin et al., 2012). In contrast, cortisol levels are naturally at lower concentrations when the body is relaxed. Cortisone is secreted at irregular pulses and is believed to be released in the last hour of sleep. Therefore, concentrations of cortisol are highest in the morning (Västfjäll, Juslin et al., 2012).

1.10.1 The Impact of Raised Cortisol Levels in People with Intellectual Disabilities

Elevated levels of cortisol are associated with cognitive impairment (Dziurkowska et al., 2021). An increasing number of studies are researching the impact of elevated cortisol levels in people with DS and other cohorts with ID. One such study explored the links between psychological stress, depression, and neurocognitive decline in patients with DS. Within this study, they found a link between reactions to traumatic events leading to an increase in the neurological responses to these events and hence accelerating the decline in cognition. In a cohort that is already at a higher risk of Alzheimer's Disease (AD), it is essential to find ways to reduce stress levels in people with DS, which could slow down the rate of cognitive decline.

An additional study by Griffioen et al. in 2023 researched the impact of dog therapy on heart rate variability (HRV) and salivary cortisol levels in 20 children with Autism or with DS (Griffioen et al., 2023). This involved taking heart rates and saliva samples before and after a six-week training programme. Saliva samples were collected before and ten minutes after the session at consistent times of the day. The researchers found that while there was no evident pattern in cortisol levels over the six –weeks, cortisol levels were significantly reduced at the end of each session compared to the recorded measure at the start.

An additional study measured the hair cortisol concentration (HCG) in adults with ID who displayed depressive symptoms before and after bright light therapy (BLT) (Hamers et al., 2021). There were 14 participants involved in the study, and the researchers found a significant correlation between the baseline levels of HCG and depressive symptoms (Hamers et al., 2021). They also found that the levels increased after the therapy sessions, particularly after dim light. The researchers believe that this is an effective measure of assessing the stress levels in cohorts with ID (Hamers et al., 2021).

1.10.2 Oxytocin and its Role in Sociability in Those with Down Syndrome

Oxytocin is a peptide produced by the hypothalamus, which is both central and peripheral to what Harvey calls actions (Harvey, 2020). Oxytocin is synthesised by the magnocellular neurons in the supraoptic nuclei (SON) and paraventricular nuclei (PVN) of the hypothalamus (Harvey, 2020). Oxytocin can also be generated in other organs, such as the kidneys, the gonads, and the pancreas (Jirikowski, 2019; Harvey, 2020).

There are variants in the oxytocin receptor (OXTR) connected to genetic makeup, resulting in individualistic responses to stress (Rodrigues, Nunes et al., 2019; Harvey, 2020). In neuroimaging studies, connectivity is evident between OXTR alleles and the right HPC, medial PFC, the dorsal anterior cingulate cortex, the amygdala, basal ganglia, and the thalamus (Harvey, 2020; Luo, Zhu et al., 2020). The OXTR is located in the orbitofrontal cortex, amygdala, HPC, the parahippocampal gyrus, and additional areas in the temporal lobe, including the anterior cingulate cortex, the hypothalamus, preoptic area and brainstem nuclei (Quintana, Rokicki et al., 2019; Harvey, 2020). Intranasal oxytocin augments the connectivity in the neural circuitry (Gordon, Jack et al., 2016). It can alter the structures of the amygdala, NAcc, insula, anterior cingulate cortex, HPC, caudate, termpero parietal cortex, dorsomedial and dorsolateral prefrontal cortex (Harvey 2020, Wu, Potenza et al., 2020).

It should be noted that oxytocin cannot be measured directly in the neurons of humans, but it is measured from peripheral fluids such as plasma, saliva, or urine. It is believed there are closer levels of consistency between concentrations of oxytocin in blood plasma and in the cerebrospinal fluid (Valstad, Alvares et al., 2017; Harvey, 2020), while concentrations in urine and saliva samples do not always reflect the correct oxytocin levels in the OXTR regions of the brain (Leng & Ludwig, 2016). Oxytocin concentrations change over time and are sensitive to activity decreasing rapidly after an event/incident (Jurek & Neumann, 2018; Harvey, 2020).

Oxytocin has traditionally been associated with lactation and muscle contraction during labour. However, oxytocin has now become associated with many social behaviours and activities. While its production is in the hypothalamus, it has been primarily associated with OXTR found in the HPC (Lin & Hsu, 2018). It helps

regulate neuronal excitability, synaptic plasticity, and is linked to social ability (Lin & Hsy, 2018). The role of plasticity has become the focus of oxytocin-based research projects (Froemke & Young, 2021). However, the significant role of oxytocin in forming social bonds, particularly in the early years, has also been emphasised (Scatliffe et al., 2019). Studies have used both plasma and saliva to ascertain oxytocin levels, making this an accessible way of assessing levels of oxytocin (Pichugina et al., 2022).

Studies have focused on its role in complex behaviours in specific cohorts, specifically those with autism spectrum disorder (ASD) and ID (Rana et al., 2023). Such behaviours include difficulties in social bonding, aggression, depression, and social recognition (Rana et al., 2023). There have been some studies that have examined oxytocin levels in children with ASD. For example, Pichigina et al. have found that children with ASD and ID had lower levels of salivary oxytocin (Pichugina et al., 2022). In an additional study by Fijioka et al. 2020, they assessed the connection between salivary oxytocin levels and attention to social information in adolescents with ASD (Fijioka et al., 2020).

While children with DS are considered very sociable, many encounter difficulties in social contexts due to cognitive impairments, particularly language barriers, social cues, and emotional regulation (Naess et al., 2016). Some studies have shown that children with DS could be more at risk than their TD peers in experiencing social functioning difficulties (Naess et al., 2016). One study by Naess et al. examined the social functioning skills of six-year-old children with DS and a non-verbal MA-matched control group (Naess et al., 2016). They found that the children with DS were less capable socially than their MA-matched peers. However, there was no difference in their ability to engage in interactive play. They found that communication skills were the predictor of difficulties with social functioning. They concluded that integrating vocabulary skills into social functioning settings at an early age would benefit these children (Naess et al., 2016). No studies have examined oxytocin levels in this cohort, which could positively contribute to finding appropriate intervention measures for children with DS.

1.11 The Neuroscience of Music

Music is an inherent part of everyday life routed in cultural identities across the globe, and music coupled with dance is the fundamental element that links all cultures and societies (Trimble & Hesdorffer, 2017). Our engagement with music sets us aside as homo sapiens, and there are biological reasons why we are more receptive to music than our ancestry apes (Trimble & Hesdorffer, 2017). This is primarily due to a more significant proportion of the brain involved in auditory processing information (Trimble & Hesdorffer, 2017). While the visual cortex is smaller in the human brain, other developments have occurred, such as an increase in the cerebellum, PFC, and the dorsal area of the temporal lobes. These are primarily associated with auditory processing, speech, and the premotor cortex, linked through the basal ganglia (Trimble & Hesdorffer, 2017).

As noted by Susanne Langer, understanding (semantics) in music came to us before meaning in language (Reichenbach, 1951; Langer, 1951; Trimble & Hesdorffer, 2017). While many enjoy music, whether performing, listening to, or singing, music's significant impact on brain structure has become a focus of attention. The influence of music on the brain was acknowledged by Charles Darwin more than two hundred years ago when they noted:

> [i]f [they] had to live [their] life again, [they] would have made a rule to read some poetry and listen to some music at least once every week, for perhaps the parts of [their] brain now atrophied would thus have been kept active through use. The loss of these tastes is a loss of happiness. It may possibly be injurious to the intellect, and more probably to the moral character, by enfeebling the emotional part of our nature (Darwin 1809–1882).

Neurological music therapy has become a growing field with a predominant role in neurorehabilitation and forms the basis of much research. Music stimulates different circuitry and, therefore, involves multisensory domains in the brain that can significantly impact those with neurological impairments (Altenmüller & Schlaug, 2012; Trimble & Hesdorffer, 2017). The role of music in therapeutic interventions has long been acknowledged. Oliver Sacks has been one of the leading figures in this area as they believed that:

> Music can lift us out of depression or move us to tears—it is a remedy, a tonic, orange juice for the ear. But for many of [their] neurological patients, music is even more—it can provide access, even when no medication can, to movement, speech, or life. For them, music is not a luxury but a necessity (Sacks, 2008).

The many ways music can impact neural pathways have also received increased interest due to the technological strides made over the last fifty years in association with the renewed emphasis on brain plasticity. It has been shown through improved imaging techniques that music can change the grey and white matter of the brain. These techniques have shown that it is possible to grow new synapses and dendrites in musical training within minutes, hours, and days (Jäncke, 2009). However, increasing grey matter, such as enlarged neurons, and synaptic density can take several weeks (Altenmüller & Schlaug, 2012). There has been much work in recent years exploring the impact of music on plasticity, such as 'Musical training induces functional plasticity in human hippocampus' by Herdener et al. in 2010. Fuki et al. also examined how 'Music facilitates the neurogenesis, regeneration, and repair of neurons' (Fukui & Toyoshima, 2008).

Studies focussing on changes in the brain in response to music and the behavioural correlates have led to the establishment of the progressive field of the Cognitive Neuroscience of Music. Zattore and Peretz's seminal publication, *The Cognitive Neuroscience of Music,* published in 2003, helped pave the way for interdisciplinary research, bringing together musicians, psychologists, and neuroscientists (Peretz & Zatorre, 2003). The correlation between brain plasticity and its effect on cognitive function has become an area with growing research (Bianco, Berchicci et al., 2022). The improved imaging techniques have revealed that many cognitive domains are affected by music. The engagement of the brain's multisensory and motor domains in response to music has, therefore, called into question the previously traditionally lateralised view of brain function.

1.11.1 Bilateral Hemispheric Responses to Music

Even just listening to music requires specific perceptual abilities, including pitch discrimination, auditory memory, and selective attention to perceive the temporal and harmonic structure of the music (Peretz & Zatorre, 2005). As acknowledged by Warren, there is no 'music centre' in the brain that can process the many contrasting

fundamental components of music, such as pitch, rhythm, timbre, and texture. The brain has traditionally been divided into symmetrical lateralisation of the dominant left and non-dominant right hemispheres, with specific functions awarded to each section. It is believed that speech and language skills are processed in the left hemisphere, while music is associated with the non-dominant right (Agrawal, Chinara et al., 2014). However, as identified by Alan Harvey, these two universal language systems involve interrelated brain structures (Harvey, 2020). Patel also believes there are correlative brain areas between music and language (Patel, 2003; Patel, 2012). These communication systems have several aspects in common, such as semantics, syntax, pitch, and rhythm (Patel, 2003). Listening to music is also believed to activate the brain's motor areas (Grahn, 2018).

In a pioneering study in Finland, led by Dr Vinoo Alluri from the University of Jyväskylä, researchers revealed how wide networks in the brain, including areas responsible for motor actions, emotions, and creativity, are all activated during music listening. This is due to the complex nature of sound and the brain areas involved in music perception. As stated by Bower, the neural networks depend on the musical task that is being perceived. Warren also believes that the musical brain is a large structure with multiple overlapping processes formed by a network of functional and anatomical links between cortical areas (Warren, 2008). The globalised approach to processing music means that music could have an interventive and therapeutic role even if a brain injury is acquired. Most data about music have been acquired by examining brain processes altered through injury or what Warren has identified as ‘amusias’. When certain musical functions are impaired while other aspects are not, this can give an insight into how music is processed in the brain (Warren, 2008).

Music is perceived via the inner ear to the brain stem and then by the thalamus, reaching the auditory cortex and the limbic centre (Weinberger, 2006). Once perceived in the auditory cortex, several regions are involved in processing. Table 1.1 shows the many affected areas.

Brain Region Affected by Music	Function
Corpus Callosum	Connects the left and right hemisphere
Motor Cortex	Movement, foot tapping and playing instruments
Prefrontal Cortex	Musical expectations and satisfaction
Nucleus Accumbens	Emotion reactions
Amygdala	Emotional reactions
Sensory Cortex	Physicality and feel for playing a musical instrument
Auditory Cortex	Listening to music
Hippocampus	Music and Memory
Visual Cortex	Reading music
Cerebellum	Movement, dancing to music and emotional reactions

Table 1.1. This table details the brain areas affected by music, as noted by Penke and Wimmer (Penke & Wimmer, 2020).

The intricate sounds the auditory cortex perceives involve pitch, rhythm, tempo, and timbre. While they are perceived as a whole, they are processed individually (Altenmüller & Schlaug, 2012; Bower, Magee et al., 2021). These sounds are managed through a complexity of cortical and subcortical areas. Rhythm is the placement of sounds in time. It is the most fundamental component of music that can exist without melody (Robertson et al., 1960). Emphasising the inherence of rhythm in human make-up, Charles Darwin stated In *The Descent of Man* (1871) that:

> it appears probable that the progenitors of man, either the males or females or both sexes, before acquiring the power of expressing their mutual love in articulate language, endeavoured to charm each other with music notes and rhythm (Darwin, 1871; Perry, 2017).

Within a rhythmic structure, there is an intrinsic beat or pulse, and the cerebellum is primarily associated with synchronising and moving to this beat or tempo. Rhythm is also believed to activate the basal ganglia (corpus striatum) and the motor and somatosensory cortices. This internal rhythm that is inherent in humans and animals has been seen in many studies. Perceiving differences in time duration tempo/beat might be linked to variations in grey matter volume in the cerebellum (Paquette, Fujii

et al., 2017). Patel's famous study with Snowball the cockatoo showed that animals also have an innate ability to synchronise to the beat but only up to 125 beats per minute (bpm) compared to humans, who can synchronise to a diversity of bpm (Patel, Iversen et al., 2009).

Pitch may be defined as a sound quality dependent on the number of oscillations. Several areas are involved in pitch recognition and memory storage. The primary auditory cortex is in the medial section of Heschel's gyrus (HG) in the superior temporal lobe. It was named after the first physician to identify the area, and it is surrounded by a complex network of cortical regions in the temporal, parietal, and frontal lobes (Warren, 2008). However, the part of the auditory cortex that plays a prominent role in pitch perception is the lateral HG in the primary auditory cortex and broodmann areas 41 and 42 (Hall & Pleck, 2009). A study by Hall and Plack found that other regions also play a role in pitch perception. They believe that the tempo-parieto-occipital junction, the planum tempora corresponding to Wernicke's area functional for language in the left hemisphere, and the PFC are involved in pitch perception (Hall & Plack, 2008).

A progression of pitches in intervallic succession creates a melodic line. Melody is believed to involve the superior temporal gyrus (STG) and parahippocampal gyri (Warren, 2008). When these melodies are familiar, they engage the anterior temporal lobe and insula (Warren, 2008). More research has also been conducted into melody and LTM and its impact on plasticity in the HPC (Groussard, La Joie et al., 2010).

When more than one pitch is presented vertically, it produces harmony. This can be presented as a homophonic or polyphonic texture that progresses horizontally in most Western music. It is believed that the areas of the brain involved in perceiving such harmony include the inferior and medial PFC, STG, and inferior parietal lobe. Such harmonic progressions are integral in creating musical syntax that involves Broca's area, which is also concerned with the motor production of speech and has a syntactical function (Maess, Koelsch et al., 2001).

Timbre (tone colour) has often been described as the 'colour' or sound quality of a tone and is often used concerning instrumental differentiation. Pitch and timbre have overlapping brain perception areas (Allen, Burton et al., 2016). However, some

studies have found that timbre is perceived in the posterior HG, the superior temporal sulcus, and the insula (Menon, Boyett-Anderson et al., 2002). It is also believed that it is processed in the temporal planum (Griffiths & Warren, 2002). A summary of the brain regions involved in music may be found in Table 1.2.

Brain Region	Musical Element
Temporal Planum **Posterior HG,** **Superior temporal sulcus, and insula**	**Timbre of voice or instrument** (Griffiths and Warren 2002) (Menon, Boyett-Anderson et al. 2002)
Lateral Part of HG	**Pitch** (Warren 2008)
Superior Temporal Gyrus	**Changes in Pitch and alterations to a melody** (Patterson, Uppenkamp et al. 2002)
Bilateral anterior portions of the temporal lobes, **Superior temporal regions,** **Parahippocampal gyri** **Insula**	**Familiar Melodies** (Satoh, Takeda et al. 2006, Warren 2008)
Inferior and medial prefrontal cortex, **Premotor cortex,** **Superior Temporal Gyrus,** **Inferior parietal lobe**	**Harmonies, Intervals and Rhythms** (Janata, Birk et al. 2002)
Broca's Area	**Musical Syntax** (Maess, Koelsch et al. 2001)
Prefrontal areas, **Cingulate cortex,** **Inferior parietal areas**	**Music as it is playing** (Särkämö, Tervaniemi et al. 2014)
Episodic memory- the hippocampus, **Medial temporal areas,** **parietal and frontal lobes** **Working Memory**	**Memory and Music** (Janata 2009) (Patel 2003, Warren 2008, Janata 2009)
Cerebellum, **Basal ganglia,** **The Motor** **somatosensory cortices**	**Rhythm, keeping the beat** (Zatorre, Chen et al. 2007, Särkämö, Tervaniemi et al. 2014)

Table 1.2. A summary of regions of the brain activated by music compiled from 'Music and the Brain: A Review of Neuroscientific and Clinical Applications' (LaRivee, 2021).

1.11.2 Far and Near Transfer of Skills: Music Training and Brain Plasticity

Music is believed to enhance many cognitive functions and is accompanied by neuroplastic changes in brain structure and function (Miendlarzewska & Trost, 2014). Brain plasticity may be defined as the capacity of the nervous system to alter its activity in response to internal and external stimuli. Over time, it can impact neuronal activity, encouraging synaptic plasticity and the brain's structures and functions (Mateos-Aparicio & Rodríguez-Moreno, 2019). Music activates many brain areas, which is evident in the brain structures of highly trained musicians, who show more remarkable plasticity in these affected areas than non-trained musicians (Miendlarzewska & Trost, 2014). Increased neuroimaging literature has evidenced this structural plasticity (Herholz & Zatorre, 2012; Barrett, Ashley et al., 2013; Miendlarzewska & Trost, 2014). For example, there is increased grey matter and functional magnet resonance imaging (fMRI) activation in the frontal, temporal, parietal, and occipital lobes, the basil ganglia, and the limbic system. There is increased white matter evident in the corpus callosum, corticospinal tracts, short fibres, and the striatum, as well as the cerebellum, which is responsible for movement (Olszewska, Marchewka et al., 2021).

Because music can impact cortical and subcortical regions, many studies have recognised its impact on other skills. The far and near transfer of skills has been reported in several studies, particularly in children. The transfer of learning is evident when a skill developed in one area impacts the abilities in another (Sala & Gobet, 2017). The near transfer of skills is considered an ability that develops in a domain similar to the original learning. In contrast, the far transfer of skills is not directly related to the original learning.

There have been many theoretical viewpoints on developing these skills, with one of the first investigations into the transfer of learning carried out by Thorndike and Woodworth (Thorndike & Woodworth, 1901; Sala & Gobet, 2017). It was Perkins and Salomon that in 1992 differentiated between the positive/negative transfer and the near and far transfer of skills. Dias in 2003 focussed on the intra-domain and inter-domain transfer of skills (Dias, 2003). The most recent research, however, focuses on how teaching methodologies can enhance this learned transfer of skills. Perkins and

Solomon, in 2012, have emphasised how critical active student engagement is in promoting this transfer of skills, which is the desired outcome of any learning (Perkins & Salomon, 2012). There has been a growth in literature in recent years promoting children's engagement with music to increase their aptitude in other subject areas and overall academic skills. For example, it has been frequently noted that music can benefit mathematics, spatial ability, and awareness (Mehr, Schachner et al., 2013). It has been said to improve general intelligence (Rickard, Bambrick et al., 2012) and literacy skills (Slater, Mohr et al., 2014). The relationship between music and language has received much attention (Patel, Iversen et al., 2009; Piro & Ortiz, 2009; Chobert, François et al., 2012) while it has also been linked to visuospatial skills (Rauscher& Zupan, 2000) and memory (Roden, Könen et al., 2014; Talamini, Altoè et al., 2017).

What skills are identified as ‘near’ or ‘far’ due to music engagement has received contrasting consideration (Sala & Gobet, 2017). A schematic representation of these near and far transfer of skills was proposed by Miendlarzewska and Trost in 2014 (see Figure 1.20) (Miendlarzewska & Trost, 2014).

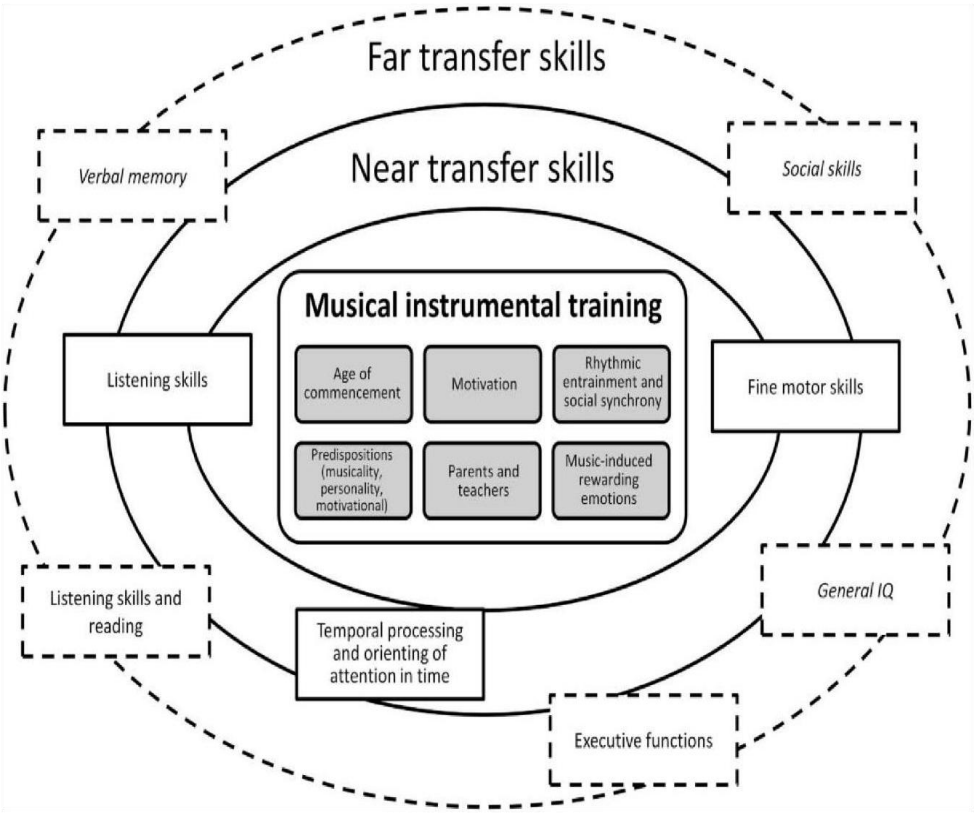


Figure 1.20. Near and Far transfer skills (Miendlarzewska & Trost, 2014). E. Miendlarzewska, & W. Trost, (2014). How musical training affects cognitive development: rhythm, reward, and other modulating variables [Review]. *Frontiers in Neuroscience*, *7*, Article 279, 1–18. https://doi.org/10.3389/fnins.2013.00279. Reproduced with permission from *Frontiers* (Creative Commons Attribution 3.0 License).

The authors identify listening skills, fine motor skills, temporal processing and orienting of attention in time as near-transfer skills impacted by music. Listening and responding are inherent aspects of musical engagement. It involves many elements, such as auditory processing, pitch discrimination and selective attention (Miendlarzewska & Trost, 2014). Developing fine motor skills is seen as a positive transfer of learning when such dexterity and fluidity of control are required to play an instrument. Music progresses in real time; therefore, WM is necessary to respond to this sequence of events (Peretz & Zatorre, 2005; Miendlarzewska & Trost, 2014).

However, the far transfer or nontrivial transfer of skills is becoming a focus of research, particularly concerning the impact of music on cognitive skills (Degé, 2021). It is proposed that if music could be proven to enhance neurological and cognitive function, policymakers might significantly emphasise music in education (Sala & Gobet, 2017). Taatgen provides an exciting framework for why such a far transfer of skills occurs (Taatgen, 2016). He believes these skills are a by-product of the learning

process that contains general cognitive skills, i.e. the skills can be quite different; however, they may activate the same cognitive pathways (Taatgen, 2016). He proposes a primitive information processing element known as the PRIMs theory (Taatgen, 2016). Central to this theory is that general cognitive skills are learned from a specifically understood task (Taatgen, 2016). For example, if a child learns the piano while practising the fingering required, it also trains selective attention, which is the cognitive by-product (Degé, 2021). These minor elements can then be built to form a PRIMs rule combined with other regulations to carry out an operator (Degé, 2021). The PRIMs rule can be used independently of the overarching skill (Degé, 2021). Therefore, an operator can be used entirely or partially for another task. This approach also addresses the early versus late transfer of skills. Taatgen outlines that when one commences learning a new skill, the transfer might be 'higher' as operators are less specific (Taatgen, 2016; Degé, 2021). Therefore, being an expert in something might hinder transfers as the operators become more specific to that particular task.

Degé points out that these links to different regions compound the idea that music lessons are perhaps associated with a domain of general cognitive ability such as EF (Hannon & Trainor, 2007; Degé, Kubicek et al., 2011). This could also account for the far transfer to general IQ (Schellenberg, 2004); literacy skills, verbal memory, and social ability that are similarly identified in Figure 1.20 as far transfer effects. However, the authors do not identify emotion in Figure 1.20 as a near or far transfer of skill. Instead, music-induced emotion is embedded in instrumental training. A body of literature shows that musically induced emotions activate areas of the brain associated with non-music-activated systems such as the amygdala, the reward system, the hypothalamus-pituitary adrenals (HPA), the amygdala, and the insula and orbitofrontal cortex (Blood & Zatorre, 2001; Miendlarzewska & Trost, 2014). Its place in the musical training strategy is significant and may seem surprising. The following sections will detail the integral role of emotion in these far-transfer skills.

1.11.3 A Neuroscientific Approach to Music and Emotions

Music has been long associated with evoking emotions. The involvement of music with feeling dates to Plato, wherein *Laws* states that "rhythm and music generally are a reproduction expressing the moods of better and worse men" (Schoen-Nazzaro,

1978). Plato also reiterated the importance of music and emotions in the *Republic.* He believed that music could evoke different kinds of feelings, such as "soberness, courage, liberality, and high-mindedness, and all their kindred and opposites, too, in all the combinations that contain and convey them" (Schoen-Nazzaro, 1978). As outlined by Peretz, there are two contrasting viewpoints on music and emotion (Peretz, Aubé et al., 2010). A 'cognitivist' believes music expresses emotions, whereas the 'emotivist' view is that music induces emotional reactions.

Music-induced emotions are related to the reward system and form one of music's most significant neurological connections. Several studies have linked the auditory cortex to the activation of the superficial part of the amygdala when listening to music (Koelsch, 2014; Liégeois-Chauvel, Bénar et al., 2014). This communication is mainly found when listening to joyful music, which Koelsch believes is enhanced by the socially elusive elements inherent in this music. The HPC has also been linked to music and emotion. Studies such as those by Trost, Ethofer et al. in 2011 and Mitterschiffthaler, Fu et al. in 2007 have noted hippocampal activation in attachment-related emotions. Music and emotion involve the NAcc through the mesolimbic pathway, also known as the reward system. The mesolimbic pathway brings dopamine from the ventral tegmental area (VTA) to the NAcc, amygdala, and HPC. The NAcc is linked to rewards, pleasure, desire, and the placebo effect (Stott & Ang, 2013).

The NAcc is activated when music is pleasurable or enjoyable. Some might describe this as experiencing 'chills' (Grewe, Nagel et al., 2007; Guhn, Hamm et al., 2007; Grewe, Kopiez et al., 2009), 'thrills' (Goldstein, 1980; Konečni, Wanic et al., 2007) or 'frisson' (Sloboda, 1991; Huron, 2006). The NAcc can also be activated by creating an 'emotional peak' where tension is made in the music through instrumentation, pitch, or rhythmic augmentation. This 'emotional peak' occurs at familiar and anticipated climactic points. An example of such an 'emotional peak' can be experienced in the transitional link between movements 3–4 in Beethoven's Symphony No. 5 in C minor Op. 67 depicted in Figure 1.21 and heard in Example One on the tracks provided. In this example, the strings play the sustained tonic of C while the bass line plays a repeated dominant note of G, whose rhythm is echoed in the timpani playing the tonic. The first violins are the only line playing an undulating

melody, enhancing the tension. The listener is held tentatively with further apprehension being introduced with tremolos in each line until the expected C arrives; however, the major key equivalent is heard rather than the expected c minor. Dopamine is the neurotransmitter released by neurons when such a reward is expected, which helps strengthen reward-related memories.
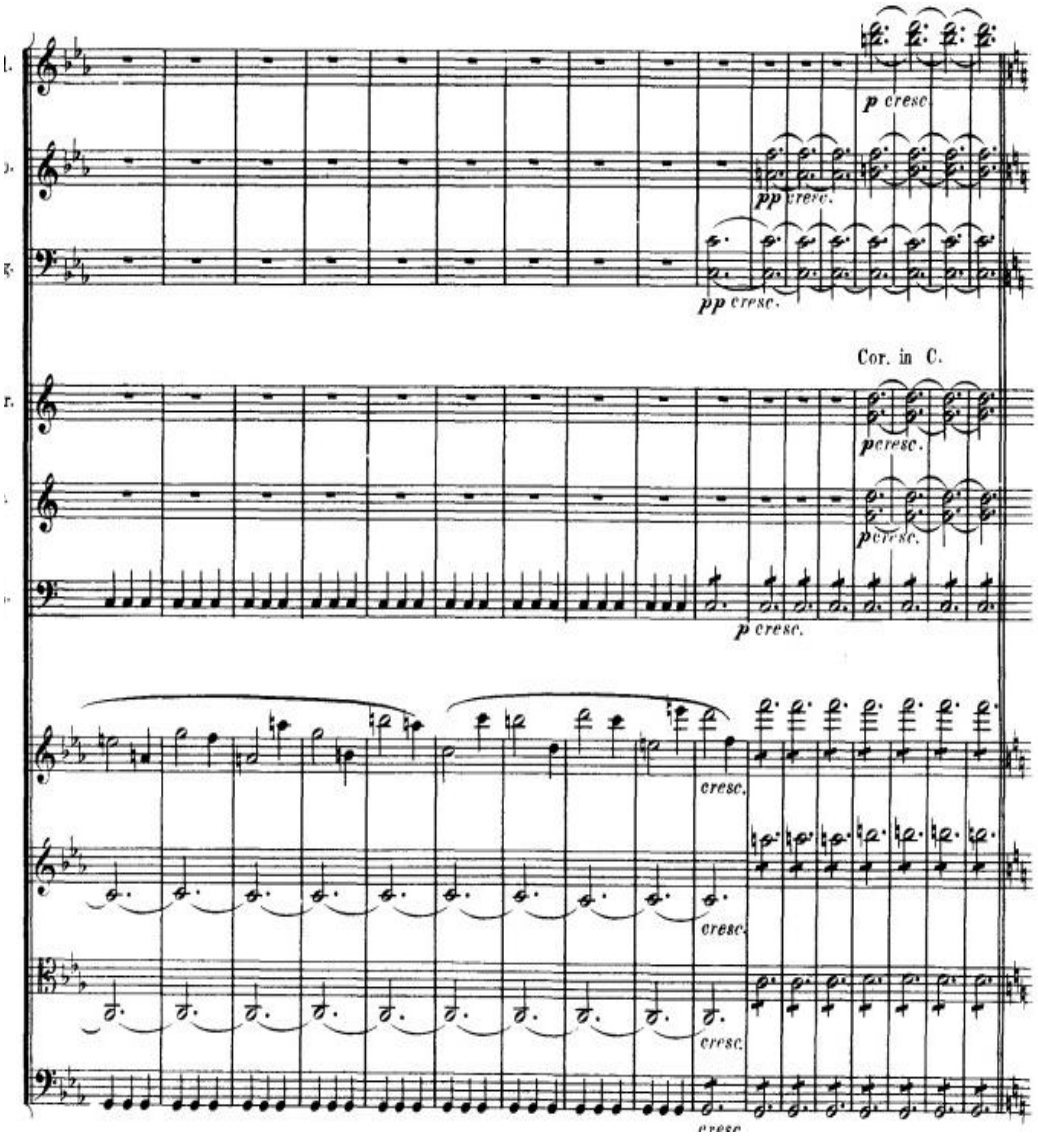


Figure 1.21. The tensional transition between Movement III Scherzo Allegro IV Allegro in Beethoven's Symphony No. 5 in C minor Op. 67. The emotional peak arrives as the piece lands in an unexpected triumphant C major, marking the arrival of movement four. Ludwig van Beethoven edited Henry Charles Litolff (1989). Symphony No. 5 in C minor Op. 67. Mineola: Dover Publications. Reproduced with permission from *Dover*.

How we respond emotionally to a selected piece of music can be influenced by the key, tempo, rhythm, dynamics, and articulation. The interconnection of music and

emotion was exploited throughout the genres by operatic and instrumental composers who strategically selected specific keys to evoke certain emotions through their music. Major keys are generally assumed to be associated with feelings of triumph and fortune, whereas more sombre and sad music is most often associated with a minor key. The triumphant C major that concludes Beethoven's Symphony No. 5 is juxtaposed with the frustration of c minor that opens the orchestral work. Coupled with tempo, selecting a particular key in music also heightens the emotive impact on the listener (Chau, Mo et al., 2016).

The influence of tempo and rhythm as cues on emotion has formed the focus of a study by Fernández-Sotos (Fernández-Sotos, Fernández-Caballero et al., 2016). It is based on a previous assumption that tempo and rhythm can regulate emotion and mood. The experiment aimed to improve the quality of life in the ageing population. Cameras and body sensors were used to monitor facial expressions and physical reactions to the music. The purpose of this project was primarily to assess the effect of note values on emotive responses. The study showed varying emotional responses in reply to the musical extracts played at either 90/120/150 bpm with specific note values. For example, when music was played at 90 bpm with semi-breves and minims, the response is relaxing, expressionless, dull, and sad. Whereas high arousal is created by music played at 150 bpm with a semi-quaver driving rhythm, which created stress (Fernández-Sotos, Fernández-Caballero et al., 2016).

Emotional responses can also be heightened with strategically placed dynamic alterations in the music. Beethoven's compositional style paved the way for dramatic dynamic changes that dominated the Romantic period. There are many examples within Beethoven's works where the composer juxtaposes contrasting dynamic levels between *pianissimo* (*pp)* and *fortissimo* (*ff*) to play with listeners' musical and emotional expectations. Referring to the tensional transition seen in Figure 1.21 and heard in Example 1, the uncertainty is intensified using a *piano* dynamic. However, the emotional intention can also be heightened when dynamics and articulation are combined, such as the chords of despair coupled with a bar of silence that brings the development section of Movement One to a close, as seen in Figure 1.22 and heard in Example 2 on the audio provided.
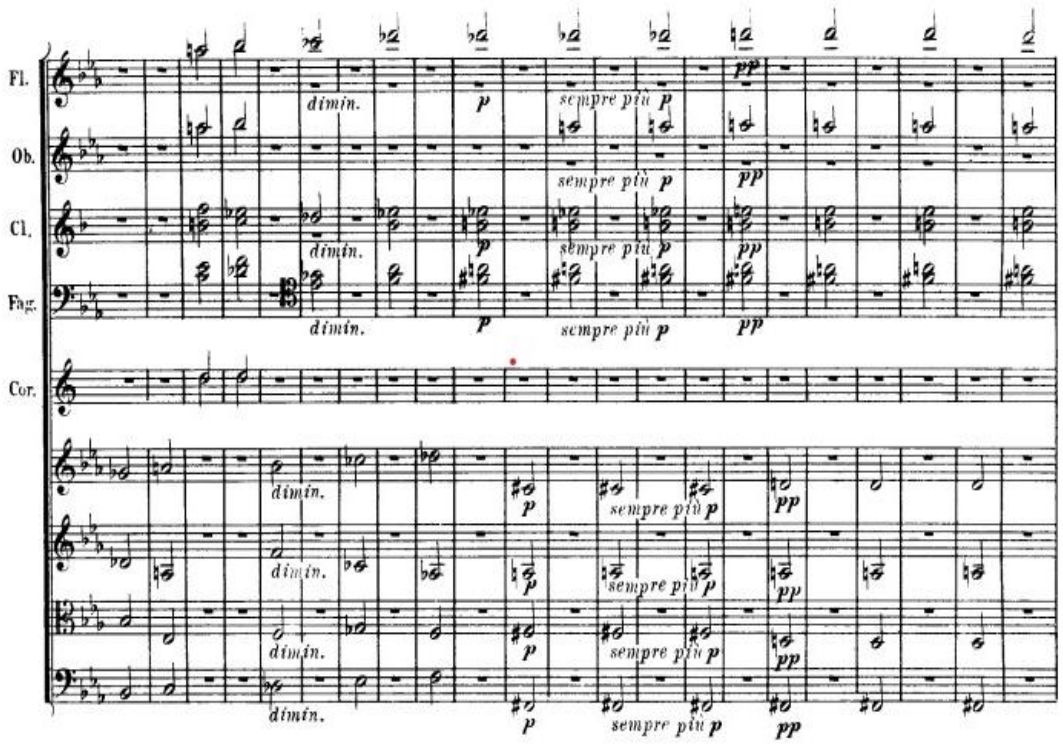


Figure 1.22. Beethoven Symphony No. 5 in C minor, Op. 67, Movement One *Allegro con Brio* at the end of the development section. Ludwig van Beethoven edited Henry Charles Litolff (1989). Symphony No. 5 in C minor Op. 67. Mineola: Dover Publications. Reproduced with permission from *Dover*.

The strategic use of harmony and harmonic progressions to heighten the impact of emotion has been employed in Popular and Western Art music for centuries. A seminal example is when a Tierce di Picardy (the Picardy 3rd) is used at the end of a minor piece. This occurs when a minor key has dominated a piece of music, yet it concludes on a major chord. For example, the well-known *Coventry Carol* is in g minor for the entire piece but ends on a G major chord to indicate the hope the newly born Christ will bring, as found in Example 3 on the audio provided. J.S. Bach frequently used the device, as did their contemporaries. As seen in Figure 1.23 and heard on Example 4, *Jesu meine Freude* BWV 81, the final chorale in J. S. Bach's Jesus *Schläft, was soll ich hoffen?* commences in a minor key yet concludes in the major key equivalent. A seminal example of the emotional effect of the Picardy third may be seen in the verse structure of *I'll Be Back* by the Beatles, in which every verse starts in a minor key and then ends with a major chord, reflecting once again the hope that 'he will be back', as heard on Example 5 on the audio provided.
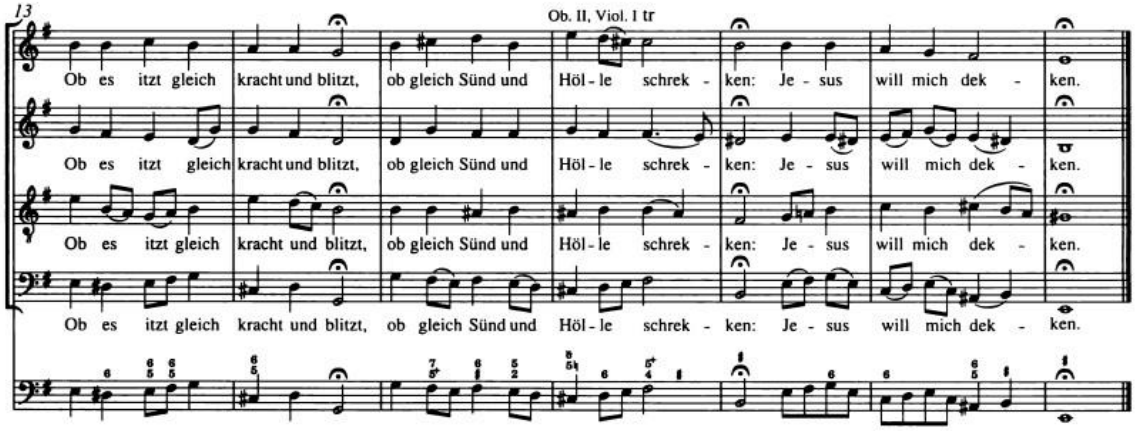


Figure 1.23. *Jesu, meine Freude* starts in e minor but ends on the chord of E major as seen in bars 13–19 from Jesus Schläft, was soll ich hoffen? BWV 81, edited by Ulrich Leisinger, Neue Bach-Ausgabe, Serie 1, Band 6, 109–36. Kassel: Bärenreiter Verlag, 1996. Plate BA 5087.

There is a distinct harmonic and melodic shift in Beethoven's Tempest Sonata in d minor Op. 31 No. 2 Allegretto (last movement). This is almost an empathetic shift in the emotion through the repetition of this opening theme and harmonic colouristic changes, as may be heard in Example 6. Similarly, there are many emotional triggers in Beethoven's Pathétique in c minor, Op. 13, No. 8 in the second movement *Adagio Cantabile*, as can be heard in Example 7.

The aforementioned musical cues invoke emotion in several diverse contexts. There is no set formulaic way of creating certain emotions, and often, they are context-dependent and reliant on several fundamental elements. However, from the above examples, there are apparent emotions, 'chills', 'frisson', or 'thrills' that can be perceived from the music. How this dynamic music affects neurochemistry will form the focus of the next section.

1.11.4 Music and the Neurochemistry of Mood

Research has shown that music can transverse all generations and cultures, leading to a bulk of research exploring the impact of music on children's behaviour toward older adults in many cultural contexts (Freeman, 2000; Tarr, Launay et al., 2014). Music significantly impacts human actions and often encourages prosocial engagement with others (Harvey, 2020). Studies have also, in recent years, acknowledged the importance of music in reducing stress levels and increasing social bonding (Thoma, Scholz et al., 2011; Thoma, Marca et al., 2013). This is partly because of its impact on the limbic system, which is connected to the reward and motivational

aspect of the brain. Within this section, the impact of music on cortisol, which is linked to stress, and oxytocin levels which is linked to social bonding, will form the focus.

(a) The Glucocorticoid Response to Music

Examining the glucocorticoid responses to music has received much attention recently due to its innate contribution to human existence on multiple levels. As seen in Figure 1.24, music has been found to impact cortisol levels in many ways, through listening to music, vocal and instrumental performance, and singing as a pastime, whether in a group or as an individual. The focus, to date, has been primarily on cortisol levels in saliva collection.



Figure 1.24. Listening to music, performing, or singing can impact cortisol levels by lowering or increasing levels, depending on the context.

(b) Rewind To Unwind: Music as a Method of Relaxation

Unsurprisingly, listening to certain types of music can help reduce cortisol levels and be a relaxation strategy (Khalfa, Bella et al., 2003). To date, many studies have explicitly focused on how 'Classical music' (Western Art Music) can reduce stress levels (Thoma et al., 2013). While it has been suggested that music and song can reduce cortisol levels in babies and children, very little quantitative data has been

conducted with such cohorts. Lim et al. recently conducted a systematic review of the literature that focuses on infant stress, specifically cortisol levels in premature babies experiencing pain (Lim et al., 2023). There have been few studies on how music therapy can reduce cortisol levels in children who are ill and need intravenous (IV) forms of medication while music is played through headphones (Hasanah, Mulatsih et al., 2020), but most of the studies carried out to date involved adult cohorts.

One of the earliest studies exploring the effect of listening to music on relaxation was carried out by Khalfa et al. (Khalfa, Bella et al., 2003). The research team recruited 24 male students to participate in the study. They were evaluated using the Trier Social Stress Test (TSST). This test includes mental calculations and public speaking in front of an audience. Saliva was collected 20–30 minutes after the participants arrived in the afternoon to serve as the baseline (Khalfa, Bella et al., 2003). They were then required to prepare a speech for 10 minutes, and saliva was collected at this stage and after the TSST. Saliva was then collected every 15 minutes for 45 minutes after the stress phase. The participants were then encouraged to relax in silence or listen to music. Nine participants in the music group listened to extracts by Enya, Vangelis, and Yanni. The results confirmed that listening to music reduced cortisol levels more effectively than silence after the primary stressor (Khalfa, Bella et al., 2003).

In a study by Thoma et al., the researchers aimed to explore the impact of music on the stress hormone cortisol and other factors (Thoma, Marca et al., 2013). Sixty females were recruited for the study, with a mean age of 25. The procedure involved an introductory welcome and an electrocardiogram (ECG) measurement followed by saliva sampling before being assigned to an intervention group. The participants were allocated to one of three groups, Relaxing Music (RM), for which the Allegri piece for double chorus *Miserere Mei* was selected. This piece has already been used in a study by Nater, Abbruzzese et al. 2006 that proved its relaxing effects (Nater, Abbruzzese et al., 2006). The second group was subjected to the sound of rippling water (SW), and the third group was not exposed to any stimulus (or rest without stimulation) (R). Saliva samples were again taken after the intervention, and salivary cortisol and salivary alpha-amylase (sAA) were measured. In addition, the researchers

measured heart rate (HR) and respiratory sinus arrhythmia (RSA). After the intervention, the participants were administered the TSST, the standardised stress test. The three conditions showed varying responses. The highest concentrations of cortisol to the stressor were found in the RM group and the lowest in the SW. However, after the stressor, sAA baseline levels were reached more quickly in the RM group than in the R group (Thoma, Marca et al., 2013).

There is the inherent belief that slow-paced music induces relaxation, whereas a piece with a quicker tempo creates excitement (Ooishi, Mukai et al., 2017). However, the findings of a study by Helsing et al. challenged this idea (Helsing, Västfjäll et al., 2016). In their research, they wanted to test their hypothesis that music would help with relaxation and, therefore, reduce cortisol and evoke positive emotions (Helsing et al., 2016). There were 41 female participants involved in the study between the ages of 25 and 45, with a mean age of 32. They were assigned randomly to either the music listening group (n=21) or the relaxation control group (n=20). The groups were required to engage with the programme for three weeks. The experimental group were instructed to pick 20 musical pieces they liked to listen to, ten relaxing and ten energising. These were then saved to the researcher's laptop, and MP3 players were supplied. In week one, the music listeners were required to relax for 30 minutes every day when they came home from work. At the end of week one, they received their MP3 players and commenced week two by listening to their selected music for 30 minutes daily. The participants then received a second MP3 player with music for week three, and they were required to listen to it every day, including weekends after work. The control group were instructed to relax for thirty minutes daily and strongly advised not to watch TV or listen to music. Many measures were encompassed in the study, including daily questionnaires and the Perceived Stress Scale (PSS), to measure stress more accurately (Cohen, Tyrrell et al., 1993). The Symptoms of Illness Checklist (SIC) and an extended version of the questionnaire were given every Thursday, as this was when the participants would collect their saliva samples. The participants were required to collect six saliva samples, three of which would be delivered every Thursday afternoon/evening and the other three on Friday morning. The Thursday samples were to be collected after coming home from work and once before and after listening to music or relaxing, depending on the

assigned group. The Friday morning samples were collected upon waking, when out of bed, and 45 minutes after waking. While there was no difference in the frequency of positive emotions between the control and experimental groups in the initial week, there was a marked increase in the experimental group in the two weeks they played their own playlists (Helsing et al., 2016). The reduced perceived stress yielded similar results, with no difference between the groups in week one. However, the PSS scores were significantly reduced in the experimental group when comparing weeks one to three (Helsing et al., 2016). There was a significant reduction in cortisol ACU in the experimental group between weeks one and three, with little or no reduction in levels found in the control group (Helsing et al., 2016). Interestingly, there was no difference between the effects of listening to energising or relaxing music.

Ooishi et al. explored the impact of contrasting tempi on the salivary levels of cortisol and oxytocin, the latter of which will be discussed in the following section (Ooishi, Mukai et al.,2017). Twenty-six males aged between 21 and 34 (mean age 29.4 years) participated in the study. Two pieces were prepared: a slow-tempo piece with 146 touches (by touches, the authors are referring to the number of times the piano key is pressed) and 56 crotchets per minute. The fast-tempo sequences had 416 contacts with a rate of 233 crotchets per minute. Each section lasted 20 minutes and included pieces by Chopin.

Musical experts were brought in to rate the pieces under six categories: tempo (0-slow to 15-fast), rhythm (0-vague to 15-outstanding), pitch level (0-low to 15-high), pitch range (0-narrow to 15-wide), harmonic complexity (0-simple to 15-complex), and consonance (0-dissonant to 15-consonant) (Gomez & Danuser, 2007; Ooishi, Mukai et al., 2017). The participants were also asked to rate their familiarity with the pieces using a scale of 0–15. In addition, self-reported questionnaires on emotional response were administered, and physiological measures such as HR, electroencephalogram (EEG), and heart rate variability (HRV) were taken. Two participants came to the laboratory at a time for two days. They were assigned either an early afternoon listening session or a later afternoon timeslot on the first day, and on the second day, they were given the same slot. The participants had a 10-minute resting period before the initial saliva sample was taken, followed by a listening session of 20 minutes, after which saliva was again collected. Heart Rate was also

monitored during the music stimulation, and a recording of their HRV followed this. The results showed an increase in oxytocin and a high-frequency element of HRV, while there was a decrease in HR when slow-tempo music was played. It was also noted that cortisol levels dropped when listening to fast-tempo music. Therefore, this study confirms that both slow and fast-tempo music can raise oxytocin levels while reducing cortisol regardless of the speed of the music.

(c) Neuropeptide Responses to Music: Oxytocin

It has been noted by several studies, such as those by Boso et al. and Koelsch, that music activates the same areas of the brain that are responsive to oxytocin (Boso, Politi et al., 2006; Koelsch, 2014; Koelsch, 2018). The studies that have focused on examining the impact of music on oxytocin, to date, can be subdivided into listening to the music, the effect on prosocial flow, and the impact of singing (seen in Figure 1.25). These will form the discussion in the sections that follow.



Figure 1.25. Oxytocin and how it promotes prosocial flow through singing and listening.

(d) Feelin' Good: Oxytocin and Music

Oxytocin is often called a 'happy hormone', as it has been identified as making us feel good, hence its connection to well-being (Ito et al., 2024). Just as cortisol levels can be lowered by listening to music, as described in previous sections, oxytocin levels have been shown to increase after listening to music. The benefits of raised oxytocin levels and their connection to music have been explored in health settings, particularly in patients undergoing surgery (Nilson, 2009).

The impact of music on postoperative patients was examined (Nilsson, 2009) based on a connection between low levels of plasma oxytocin, pain and anxiety levels (Evans, 1997). Forty participants in the study had either undergone an aortic valve replacement (AVR) or a coronary artery bypass grafting (CABG) and were prescribed 12 hours of bed rest. Twenty patients were assigned to the group that listened to music the day after the operation, while the others recovered without music (Nilsson, 2009). The music group were given an MP3 player, which they heard through a pillow. The music consisted of 60–80 bpm melodies at a volume of 50–60 decibels for thirty minutes. Plasma oxytocin, HR, mean arterial blood pressure, partial pressure of oxygen (PaO2) and arterial oxygen saturation (SaO2) were measured. The oxytocin levels in those with the music intervention were significantly higher than those in the control group (Nilsson, 2009).

Unlike cortisol, Oosishi, Mukai et al. found oxytocin responses to be more sensitive to some of the fundamental components of music (Ooishi, Mukai et al., 2017). Interestingly, Ooishi, Mukai et al. found a reduced HR and increased oxytocin level when listening to slow, relaxing music. Still, when a faster tempo music was played, oxytocin levels remained the same if slightly reduced. In contrast, cortisol levels rose slightly (100pg/ml) after listening to slow music. A significant decrease in cortisol levels (200pg/ml) was recorded after listening to fast-tempo music.

It has been found in some studies that listening to 'sad' music can be as pleasurable as listening to 'happy' or more upbeat music (Eerola, Vuoskoski et al., 2021). As stated by Eerola et al., the challenge of examining the physiological effect of music is because much of this is considered 'sad' music, which is linked to memories or poignant lyrics. In one study, researchers wanted to test the impact of

sad and unfamiliar music (Eerola, Vuoskoski et al., 2021). Sixty-two women engaged with the research and either listened to 'sad' music or no music and were recruited based on high and low empathy for 'sad' music. The music was selected based on previous studies proving that specific music pieces evoked sadness. These included the second movement of Mozart's *Piano Concerto No. 23*, *Les Siècles*; *Discovery of the Camp* by Michael Kamen, and *Oblivion by Astor Piazzolla* (Vuoskoski & Eerola, 2012; Eerola, Peltola et al., 2015; Eerola, Vuoskoski et al., 2016). The participants were seated in comfortable chairs, and blood was taken to measure prolactin (PRL), oxytocin, cortisol and adrenocorticotropic hormone before exposure to the music. The participants were allowed to listen to music or remain silent for 20 minutes, and blood samples were again taken. This was followed by another 20-minute session in which participants were exposed to the opposite condition as previously experienced. Prolactin concentration was lower after listening to music. This is consistent with the results from other studies that state that listening to music activates the dopaminergic reward system, inhibiting the secretion of PRL. Interestingly, there was a decrease in oxytocin in the highly empathetic group but an increase in oxytocin in the less empathetic group (Eerola, Vuoskoski et al., 2021). This indicates that individualistic sympathetic responses to music can influence the endocrine system.

(e) Oxytocin and the Prosocial Flow of Song

While some studies have shown adverse effects of increased oxytocin levels, such as increased competition and aggressive tendencies in males, the prosocial impact of increased oxytocin dominates results, particularly in relation to music (Fischer-Shofty, Levkovitz et al., 2013; Fischer-Shofty, Shamay-Tsoory et al., 2013; Alcorn, Green et al., 2015; Harvey, 2020). It has also been shown to indicate well-being, as reflected by Suzuki et al., who found reduced salivary oxytocin levels in maltreated children (Suzuki, Fujisawa et al., 2020). Lullabies sung by mothers to their infants are believed to increase salivary oxytocin and reduce cortisol to improve well-being and reduce postpartum depression while increasing social bonding (Wulff et al., 2021). There have been invaluable studies carried out by Strathearn et al. 2009 that explore the increased oxytocin in mothers who interact with their babies and bond with

them. Other studies have shown the potential relationship between music and heightened oxytocin levels in the context of sociability (Keeler, Roth et al., 2015). However, as described in the previous section, studies have focussed more on older age profiles when examining oxytocin levels and music.

In a study by Keeler et al., the researchers wanted to examine how musicians and people listening to music often talk about getting lost in the music (Keeler, Roth et al., 2015). The authors make an interesting connection with the psychological state of 'flow' where someone is immersed in a task (Csikszentmihalyi, 1975; Keeler, Roth et al., 2015). Csikszentmihalyi first explored this sense of 'flow' experience and described it as "the holistic sensation that people feel when they act with total involvement" (Csikszentmihalyi, 1975). The 'flow state' experience through music has also been explored by Chirico et al. (Chirico, Serino et al., 2015). It is believed that when performed as a group, this 'flow state' can lead to interpersonal connections (Keeler, Roth et al., 2015). This is significant as many cultures have distinct music that relies on group performance, such as Irish Traditional Music (Keeler, Roth et al., 2015; Hart & Blas, 2015).

The role music plays in promoting social bonds has only grown in emphasis since the latter part of the 1980s (Bowling, Gahr et al., 2022), with the most recent focus documented by Savage et al. and their Music and Social Bonding (MSB) (Savage, Louis et al., 2020). Synchronicity also promotes such bonding (Spengler, Scheele et al., 2017)—this can be experienced through drumming activities but, most often, through group singing (Mogan, Fischer et al., 2017).

While the impact of instrumental playing on oxytocin levels has received some attention, group singing has been the predominant focus (Kirschner & Tomasello, 2010; Minutillo, Panza et al., 2020). Its prominence has been due to the prosocial impact of group and choral singing and its connection to well-being and increased social bonding, particularly in older adults (Grape, Sandgren et al., 2003; Clift, Nicol et al., 2010). It is believed that singing may, in fact, have evolved to accommodate such social bonding (Dissanayake, 2000; Kreutz, 2014). The tradition of choral singing has been examined in many contexts. In a study by Fernández-Herranz, Ferreras-Mencia et al. 2021, that included 1,513 participants, they found that singing as a

group can create a sense of belonging and increase social interaction on many levels (Fernández-Herranz, Ferreras-Mencia et al., 2021).

Choirs and singing groups can act as supportive structures for people with fewer opportunities to interact socially; hence, there has been a considerable increase in the number of choirs established to support those with dementia or Parkinson's and their carers, for example. In recent years, there has been an increase in the number of choruses and vocal ensembles that have contributed to such studies to understand the neurochemical processes involved in this prosocial bonding that occurs when singing as a group.

Bowling et al. have conducted a research project that explored the impact of group singing on cortisol, oxytocin and testosterone levels (Bowling, Gahr et al., 2022). Like many other research projects, this has been influenced by studies, such as that performed by Cohen et al. in 2006, which examined the impact of group singing on the psychological well-being of participants. However, as noted by Bowling, studies have shown both increases and decreases in oxytocin levels in participants depending on the context in which they were singing. For example, a study by Keeler and colleagues (2015) tested plasma oxytocin and ACTH before and after two contrasting singing conditions, improvised and pre-composed. These assessed social affiliation, arousal, and engagement (Keeler et al., 2015). They found significant decreases in ACTH, specifically in the pre-composed conditions. Oxytocin levels were raised only in the improvised singing condition (Keeler et al., 2015). This could be due to the altered activity in the PFC, and inevitably, the nature of the activity enhanced the prosocial interaction. As acknowledged by the researchers, further investigatory projects with larger test samples are required to understand the full implications of improvisatory singing.

However, Fancourt et al. (2016) found decreased levels of salivary oxytocin in their study, where they assessed the impact of group singing on mood, immune response, and stress in patients with cancer diagnoses. Similarly, Schladt 2017, researched the effect of solo versus group singing on oxytocin and cortisol levels before and after each activity over an intervention period. While cortisol levels were reduced in both conditions, salivary oxytocin was increased after solo singing (116%

of basal), whereas the levels were significantly reduced after the group singing (81% of basal) (Schladt, 2017).

Forty-five females and 26 males, aged between 17 and 28 years, were involved in Bowling's study. The study occurred mid-way through the winter semester when the singers had been singing together for eight weeks. There were four conditions: 1) speaking together, 2) singing together, 3) speaking alone, and 4) singing alone. The rehearsal occurred between 18:00 and 20:00 hours on a Monday evening. Saliva collection and questionnaires were collected before a 20-minute core activity. The speaking and singing activities were carried out in two different rooms. The choir director led the rehearsal, with 5 minutes dedicated to warm-ups and 15 minutes for repertoire rehearsal. In the speaking exercise, a choir member led the group in articulating the lyrics of the same songs for the same duration in no particular rhythm. The project found decreases in cortisol levels, 29% in those singing together and 31% in those singing alone. The study also noted that oxytocin decreased after singing, which the researcher believes was a direct result of working with an existing choir, as the participants had current friendships and knew each other. In contrast to an 'increase study' by Kreutz et al., the researchers worked with a purposely created choir; therefore, the social interaction and bonding were more explicit (Kreutz, 2014).

Good et al. used a sociobiological approach to assess the mood-boosting effect of singing both in a group and individually (Good & Russo, 2021). The researchers tested salivary oxytocin and cortisol before and after the task, and a self-report mood assessment was performed, as well as positive and negative affect schedules (PANAS) (Watson, Clark et al., 1988). Focusing on older adults, nine participants agreed to participate in the study, with a mean age of 72.8 years. The Montreal Cognitive Assessment (MoCA) recorded all participants as having no cognitive incapacities. Saliva was collected immediately before and after the 45-minute sessions. The group singing included breathing exercises, warm-ups, and songs (Good & Russo, 2021). However, interestingly, the individual singing was a re-creation of a choir director leading a choir through activities. This study showed elevated levels of salivary oxytocin in participants after the group singing activities compared to those who participated in individual singing sessions. In contrast,

cortisol levels were reduced, consistent with evidence that dance and singing activate oxytocinergic networks (Harvey, 2020). The researchers also found a correlation between mood and increased oxytocin levels, with no correlation between cortisol and mood. However, this raises important questions about environmental conditions and the significance of low/high stress performing conditions impacting neurochemistry (Good & Russo, 2021).

Considering the above studies, the following sections will focus on the role of music as an interventional strategy and how it has been used with younger children.

1.12 Music as Intervention

An intervention is "a planned and systematically implemented activity in current social structures, which aims at changing knowledge, attitude or behaviour of a person, an organisation, or a population" (Loos, 2008). When approaching a literature review on the implications of musical engagement on children, the language used can give an unbalanced reflection on the kinds of music programmes being delivered. For example, if the search includes the word intervention, the results will be specifically related to therapeutic interventions for those with additional needs. Therefore, the search had to be broadened to find music programmes with TD groups. However, as this section unfolds, it will become evident that 'interventions and interventional strategies' are applicable and used in all areas of society across all demographics, even if the terminology used differs.

1.12.1 A Question of Intervention: Music with Typically Developing Children

The intervention strategies (or otherwise called) have contrasted considerably in the music activity, methodologies used, and the dosage or amount of exposure within a music intervention. The following section will explore some areas and strategies used in these studies that have been carried out with the TD population.

These contrasting approaches were examined in a systematic review by Dumont et al. (Dumont, Syurina et al., 2017). In this study, they considered 46 articles extrapolated from four databases. The articles reviewed primarily focused on 6–13-year-olds, with varying sample sizes within these studies. The selected papers showed a diversity of musical approaches incorporated in these interventions,

including various instruments, song singing, listening, composing and an understanding of music fundamentals, as seen in Figure 1.26.



Figure 1.26. The number of methods included in intervention strategies is highlighted by Dumont et al. in their systematic review (Dumont, Syurina et al., 2017).

The reviewers categorised the intervention strategies and outcomes into motor skill development, social and emotional development, language, cognitive development, academic performance, and what they classified as non-musical-related skills, as seen in Figure 1.27 (Dumont, Syurina et al., 2017). They also discovered that the dosages used in these interventions varied considerably, from two days a week to daily, while the duration also varied. The differentiated approaches will form the focus of the following sections, exposing the considerable variations in adopted practices in these studies.



Figure 1.27. The focus of intervention studies, to date, has been on developing motor, cognitive, social and emotional skills. As highlighted by (Dumont, Syurina et al., 2017), academic, language and non-motor skills have also been considered.

(a) Motor Skills: Instrument, Music and Movement, Handclapping

Music engagement has been chiefly associated with instrumental playing and the development of fine motor skills. For example, Costa-Giomi explored how playing the piano for two years impacted the fine motor skills of children who had never engaged with an instrument before compared to those who did not receive piano tuition (Costa-Giomi, 2005). There was a significant improvement in the fine motor skills of those who received the lessons and the speed at which they responded to specific stimuli (Costa-Giomi, 2005).

Many researchers have explored using music to enhance gross motor development in early childhood. For example, Zachopolou et al. conducted a study involving ninety children. Fifty children were involved in a music and movement programme for two months, while the control was involved in a physical education programme. On completion, the children involved in the music and movement programme dramatically improved in jumping and dynamic balance (Zachopolou et al., 2004).

Janzen et al. also conducted a study that evaluated whether 2 hours a week of music activities would improve movement against a control group. The results found that those who engaged with music had considerably more precision in discrete and continuous movement (Braun et al., 2014). More recently, Ul Badriyak et al. examined the impact of 'Gross Motor Ability in Early Childhood Through Motion and Song Activities' (Badriyah, Ristyadewi et al., 2020). Within this discussion, they emphasise the importance of song and movement to enhance these gross motor

skills, enabling the child to become familiar with themselves and heighten awareness of their surroundings (Badriyah, Ristyadewi et al., 2020).

Handclapping songs involve sensory-motor actions, including singing, hearing, touching and motor skills (Brodsky &Sulkin, 2011). It also involves social interactions between partners that require verbal interactions (Brodsky & Sulkin, 2011). Therefore, in three contrasting studies, Brodsky and Sulkin considered the impact of handclapping songs and its potential effect on cognitive and motor skill development in children aged 5–10 (Brodsky & Sulkin, 2011). The first element assessed the relationship between handclapping to music and academic achievement in 18 children with a mean age of 7. This involved the rote learning of two handclapping songs over three weeks. The results revealed that those proficient in handclapping to songs were also more efficient learners. The second experiment included ten children aged between 8 and 8.5 years. This experiment involved bimanual rhythmic patting and aural dictation. Five participants reported handclapping to songs, whereas the other five did not. The five who spontaneously engaged in handclapping activities were more proficient at aural dictation and had better eye-hand motor sequences (Brodsky & Sulkin, 2011; Dumont, Syurina et al., 2017). In the third experiment, 27 children were engaged in a handclapping intervention for eight weeks, while 24 received a music appreciation guided listening curriculum (MAGL) for the same duration. Those who received the handclapping intervention found handwriting more accessible than those who did not (Brodsky & Sulkin 2011, Dumont, Syurina et al. 2017). Therefore, it can be concluded that handclapping can positively impact cognitive skills and motor development.

(b) Music and Language Acquisition

The inherent links between music and language have long-established connections, with some detailed research found at the neurological and cognitive levels. In their review, Dumont et al. noticed two predominant approaches to finding intrinsic links between music and language (Dumont, Syurina et al., 2017). Firstly, studies tend to focus either on phonological awareness and auditory processing or the development of reading skills. These can be divided into subareas focusing predominantly on younger children (Vidal, Lousada et al., 2020).

Patel believed in such an intrinsic link between music and language that he devised the OPERA hypothesis. Within this theory, he believes there is an overlap between parts of the brain when responding to music and hence believes that music training enhances the neural encoding of speech (Patel, 2011). The plasticity in speech networks is met when the following conditions occur: **O**verlap, **P**recision, **E**motion, **R**epetition and **A**ttention. By **O**verlap, Patel refers to the intersecting anatomical regions in the brain responsible for music and speech. The **P**recision required for music is more demanding than speech in these shared regions and, therefore, could assist in strengthening pathways in the areas needed for speech. As seen in a previous section, **E**motion is inherent in music and helps to create positive emotional responses. The **R**epetition that is so often found in music again reaffirms these neural pathways. Finally, focused **A**ttention, which is associated with music, is beneficial for speech encoding as music places higher demands on neural processing systems. Therefore, Patel hypothesises that OPERA processes benefit speech acquisition (Patel, 2011).

Escalda et al. conducted a study with 56 children exploring the relationship between music experience, phonological awareness skills, and auditory processing abilities (Escalda, Lemos et al., 2011; Dumont, Syurina et al., 2017). The results showed that the 26 children with music experience performed significantly better on auditory awareness processing and phonological skills than the 30 children who did not (Escalda, Lemos et al., 2011; Dumont, Syurina et al., 2017). Most recently, Vidal et al. explored this further in a study with forty-four children aged 3–4 (Vidal, Lousada et al., 2020). The researchers used a pre- and post-test design. Their phonological awareness was tested before a weekly music intervention or a visual arts programme for a school year. There was a general improvement in both groups due to developmental progression; however, the music cohort showed significant improvement in phonological awareness (Vidal, Lousada et al., 2020).

While numerous studies have examined the relationship between music and reading skills, many results have been inconclusive (Cogo-Moreira, Brandão de Ávila et al., 2013; Dumont, Syurina et al., 2017). Further studies are required to establish music's impact on speech and language acquisition.

(c) The Impact of Music on Social, Emotional Skills and Wellbeing

Inspired by a publication by Weinberg and Joseph, this section explores the interventional strategies that have been applied to assess the impact of music on well-being and, specifically, its real implications on social and emotional skills (Weinberg & Joseph, 2017). Ritblatt et al., for example, found that a music intervention in children aged between 3–7 years impacted their socioemotional skills, as reported by the teachers, compared to those who did not (Ritblatt, Longstreth et al., 2013). Schellenberg et al. saw increased sympathy and prosocial behaviour in those immersed in a music pedagogical programme (Schellenberg, Corrigall et al., 2015). Kirschner and Tomasello conducted a study with forty-eight 4-year-olds who were put in pairs and assigned to either an episode of interactive play with music or an interactive play session without music (Kirschner & Tomasello, 2010. The music condition showed increased interactivity between the participants when confronted with a problem to solve (Kirschner & Tomasello, 2010). However, in contrast, a study by Rickard et al. in 2013 reported no evident benefits to children aged 5–8 years who received a specialised music education under their school's existing music education system (Rickard, Appelman et al., 2013).

A recent study by Diaz Abrahan and colleagues discovered that musical improvisation over music reproduction could potentially improve social skills and memory (Diaz & Justel et al., 2022). While using improvisation as an intervention tool is interesting in this context, the specific details of such music interventions are rarely specified. Similarly, studies examining music's impact on emotional and social development have mixed methods and findings (Schellenberg & Mankarious, 2012; Rabinowitch, Cross et al., 2013).

(d) Intelligence, Memory, and Attention.

The connection between music and IQ potential has been the central topic of recent publications. Schellenberg was the first to study the implications of music engagement on IQ (Schellenberg, 2004). Within this study, 144 children were divided into four groups: two receiving music, one engaging in vocal lessons and the other in keyboard tuition, and two control groups, one receiving drama while the other group

received no lessons. The results showed a slight increase in IQ in those who received music and drama lessons.

Kaviani and colleagues randomly assigned sixty 5–6-year-olds to either a group receiving music lessons based on the Orff music programme or a group not receiving lessons (Kaviani, Mirbaha et al., 2013). After three months and using the Stanford-Binet Intelligence Scale, the researchers found that the music group had significantly better STM, abstract visual reasoning, and verbal reasoning (Kaviani, Mirbaha et al., 2013; Dumont, Syurina et al., 2017).

An additional study by Valesco tested the musical aptitudes of primary school children in relation to their IQ (Valesco, 2017). Eighty-five students aged between 9 and 13 participated in the study. This project revealed a relationship between IQ and musical skills. It also showed that the higher the IQ, the higher the musical ability. Finally, it demonstrated a difference in IQ depending on age, indicating the importance of CA-matching and MA-matching in further studies (Valeson, 2017).

In a computerised music training intervention by Moreno et al., researchers found enhanced memory for vocabulary in those who engaged with the music training programme compared to those who received visual art training (Moreno, Friesen et al., 2011). Bugos and Jacobs conducted a study examining the impact of Composers in Public Schools (CiPS) with 28 children (Bugos & Jacobs, 2012). Of these participants, 15 students were assigned to the music programme, while 13 were assigned to the control group. The researchers found that the music group showed enhanced performance in the Wechsler Intelligence Scale for Children, fourth edition (WISC-IV), but did not see any vocabulary improvement. While some impact has been found on IQ, these studies show much diversity in approaches. More consistently designed studies are needed to ascertain the effects of music on IQ.

Recent studies have indicated that music training can improve memory (Talamini, Altoè et al., 2017; Mansens, Deeg et al., 2018; Harvey, 2020). The influence of music on memory has received considerable attention in adults, specifically older adults (Rodrigues et al., 2021; James et al., 2021). While fewer in number, there have been, however, several studies that have focussed on children. A longitudinal survey by Degé et al. found that an extended music programme that spanned over two years significantly impacted short-term visual and auditory memory scores (Degé, Kubicek

et al., 2011; Dumont, Syurina et al., 2017). Similarly, Roden et al. found that children who received a school-based music programme outperformed those with a natural science curriculum on verbal memory (Roden, Kreutz et al., 2012). After a handclapping intervention, Brodsky and Sulkin showed improved verbal memory (Brodsky & Sulkin, 2011). In 2017, a study was carried out by Ginting et al., entitled 'Improving the Memory through Singing Method of Children Ages 5–6 Years in Kindergarten Insan Pandhega' (Ginting, 2019). This study aimed to assist children with retaining their parents' full name, address, and parent's phone number. At the study's conclusion, the research revealed that music could be a strategic tool to assist memory in young children. The children in the 'well-developed' to 'very well-developed memory' category increased from 36% to 80%.

(e) Executive Functions and Music

Loui and Guetta (2019) believe music involves top-down and bottom-up processes that require attention and EF. The role of music in EF is an aspect of cognitive function that is important in this current research project. In a study by Kahlil et al., the researchers examined the impact of music, specifically group synchronicity, on attention in children with attention deficit hyperactivity disorder (ADHD) (Khalil, Minces et al., 2013). This study showed increased attention and fewer symptoms of ADHD in students aged 7–12 who could synchronise to a rhythmic beat. Similarly, Zuk et al. (2014) found that children who played a musical instrument for an average of 5.2 years performed better on cognitive flexibility tasks and processing speed tasks than those who did not play an instrument (Zuk, Benjamin et al., 2014).

In a more recent research project by Janurik et al., they examined the relationship between music perception and EF (Janurik, Szabo et al., 2019). Interestingly, they investigated 'simple music activities and their role in the developmental trajectory of EF in this cohort' (Janurik, Szabo et al., 2019). There were 131 participants involved in the study with an average age of 7 years and two months. Improvements in pitch and time perception impacted EF significantly. In an additional study, Shen et al. examined the impact of music training on EF in preschool children (Shen, Lin et al., 2019). There were 61 children involved in the study from a university-affiliated preschool in North China. The three components of EF examined were

inhibitory control, WM, and cognitive flexibility. While the control group were involved in usual classroom activities, the experimental group were engaged in a 12-week music programme that included music theory, singing, dance and role play. Executive Functions were tested before (T1) and after (T2), while the effect of the music programme was assessed after a cessation period (T3). It was found that music training can promote EF even after the intervention period.

Working memory has also been the focus of several studies. For example, one longitudinal study by Roden et al. examined WM in 25 children aged 7–10 (Roden, Könen et al., 2014). Some children engaged with an extended music instrument programme, while others engaged with a science programme. The researchers found that there were positive impacts on WM. In support of this finding, Portowitz et al. also saw increases in children's WM scores after engagement with the In Harmony music programme supported by new technologies (Portowitz, Peppler et al., 2014).

1.12.2 Music and Individuals with Down Syndrome

The research that has been carried out to date has highlighted the potential benefits of musical engagement on behavioural and academic performance in TD cohorts. However, while music therapy has a detailed history of being used with those with additional needs (Rushton et al., 2023), the benefits of music on the development of those with DS have seen very little research both from a music training perspective and in therapeutic areas. In a comprehensive article published in 2020, Gemma et al. presented a systematic review of the specific studies exploring music's impact on children with DS (Gemma, Pablo et al., 2020). This section will analyse this review to expose the lack of detailed research that has been carried out, the methodologies used, and the specific outputs. While Gemma et al.'s review will form the focus of the following section, additional more recent articles that were not included in the review will subsequently be discussed.

As reiterated by Gemma et al., several music and music education studies report the importance and benefits of music in the lives of children with disabilities. The authors focused on studies published from 2002 onwards that included populations with DS, excluding non-peer-reviewed studies such as dissertations. While the content of the music interventions used overlap, the aims of these projects

were very different. Therefore, the main areas studied were motor development, communication development, social and emotional development, cognitive development, and multi-area studies that dealt with more than one development area. The breakdown between each region may be seen in Figure 1.28 (Gemma, Pablo et al., 2020).

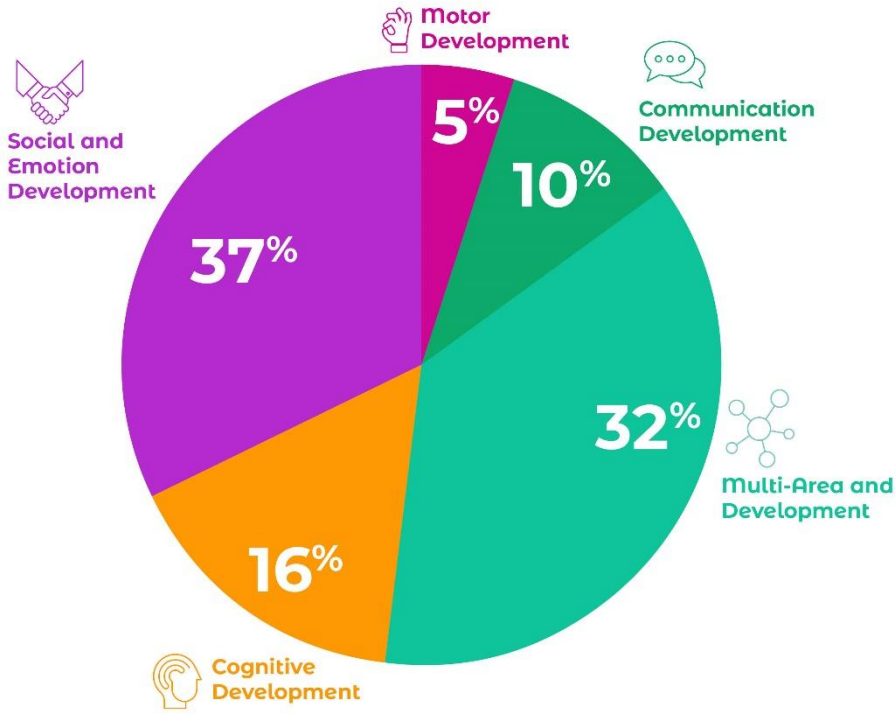


Figure 1.28. This visual representation was created from the systematic review by Gemma et al. It shows the statistical breakdown between motor skills, social and emotional development, communication development, as well as the multi-areas of development and cognitive domains (Gemma, Pablo et al., 2020).

Interestingly, Gemma et al. note that 11 studies were published after 2010, while eight were published between 2002 and 2009. Only 10/19 articles focused specifically on those with DS. In seven articles, the sample included several patients with complex psychological and motor disorders, while two studies compared DS and TD cohorts. Five studies focused on participants and their families, while another two reviewed previous research projects. The breakdown of the contents of these articles may be seen in Figure 1.29.

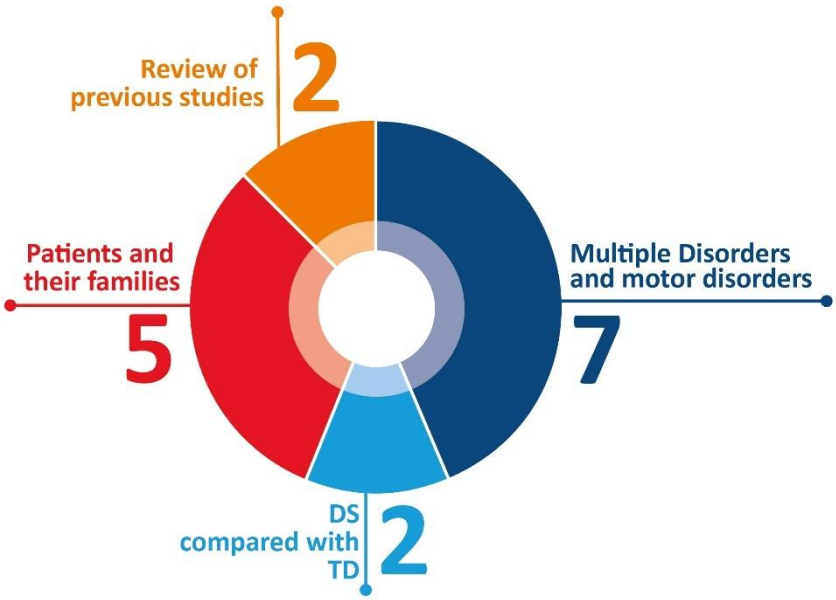


Figure 1.29. A breakdown of the contents of ten articles that addressed the impact of music on people with DS (Gemma, Pablo et al., 2020).

(a) Fine and Gross Motor Skills and Music

The importance of music and its impact on motor skills in the TD population has been previously documented; however, only 5% of the studies identified by Gemma et al. emphasised its importance in people with DS. As will be reflected, much of the research on music with DS cohorts has been scant, with considerable disparities in experimental design (Guy & Neve, 2005). Previously, studies have focused on addressing this cohort's gross and fine motor skills; a summary of these may be seen in Table 1.3.

Camps, Prina, and Rodriguez conducted a quasi-experimental study of two groups of four children with DS. Within this study, the researchers combined a 'psychomotor session' with music, which aimed to improve the gross motor skills of these children, specifically coordination, spatial organisation, and balance. However, the results were inconclusive, with huge disparities within the experimental group (Camps et al. 2017, as found in Gemma, Pablo et al., 2020). Gemma et al. also refer to a study by Becker and Dusing that included 14 weeks of dance (jazz and ballet), voice and acting to increase the children's ability to run effectively. At the end of the study, there was a significant improvement in balance and the ability to run at

increased speed. In an additional study, parents were interviewed after an intervention that included music therapy-based games; they reported improved motor skills (Pineda & Pérez, 2011).

ELEMENT	AFFECTED AREA	AUTHOR
Gross Motor Development	Co-ordination	Becker and Dusing (2010) Caps, Prina and Rodriguez (2017)
	Balance	Caps, Prina and Rodriguez (2017)
	Spatial Organisation	Becker and Dusing (2010) Caps, Prina and Rodriguez (2017)
Fine Motor Development	Fine motor skills Oral motor skills	Guy and Neve (2005)
	Motor skills in general	Pineda and Pérez (2011)

Table 1.3. The motor skills affected by music in those with DS were compiled from the review by Gemma et al. (Gemma, Pablo et al., 2020).

(b) Communication and Language Skills

The atypical development of communication and language skills in children with DS is one of the prevailing characteristics of this cohort. As identified by Gemma et al., only 10% of the studies examined or explored this aspect within their intervention. However, while limited, the studies acknowledged the importance of verbal or nonverbal interventional strategies to assist communication and language development. Pienaar's qualitative study, for example, involved 19 caregivers working in schools specifically for those with additional needs (Pienaar, 2012). This study aimed to examine the effects of music therapy sessions on children from the caregivers' perspectives. Through an anonymous questionnaire, the study concluded that the caregivers noted that music therapy sessions effectively encourage communication through music interaction (Pienaar, 2012). They also saw the potential this intervention strategy of making sounds, singing songs, and playing

musical instruments could have on the language development of children with DS (Pienaar, 2012). Mizuno and Sakumo found similar findings in their study that assessed the impact of playing a drum on children with disabilities. They found that playing a wadaiko drum (traditional Japanese drum) could impact the communication skills of these children (Mizuno & Sakuma, 2013). Throughout a 14-week intervention, they found that this nonverbal communication facilitated shared gestures and eye contact through rhythm. Guy and Neve similarly recognised the impact of music engagement on communication skills and language. Their study aimed to examine if using a simple musical instrument, such as a slide whistle, could strengthen the development of orbicularis oris muscles that could, in turn, assist language development. The researchers also emphasised the need for collaboration between SLTs and music therapists to optimise communication realisation (Guy & Neve, 2005).

Vitoria-Gallastegi conducted an experimental programme for students with additional needs over three months. By providing music education to these students, the researchers aimed to assess the impact of music on their psycholinguistic abilities. After the study, the researchers found a link between musical experience and improved auditory music processing, verbal expression, and memory (Vitoria-Gallastegi, 2005). This was one of few studies to use standardised tests to assess psycholinguistic abilities (Gemma, Pablo et al., 2020).

More recently, Pineda and Pérez conducted a study to explore the impact of a 9-month music therapy programme on language development in a cohort of children with DS. The researchers found the children showed more extensive vocabulary development and improved pronunciation of phonemes (Pineda & Pérez, 2011). A summary of the implemented strategies may be found in Table 1.4, and the breakdown between verbal and nonverbal components can be seen in Table 1.5, all of which were believed to contribute to enhanced thoughts and feelings (Gemma, Pablo et al., 2020). The importance of non-verbal communication is promising and should be considered in future intervention studies and their design.

ELEMENT	AFFECTED AREA	AUTHOR
Communication	Repeated Sounds	Pineda and Pérez (2011) Pienaar (2012)
	Synchronicity	Mizuno and Sakuma (2013)
	Communication skills, more extensive vocabulary and enhanced pronunciation of phonemes	Pineda and Pérez (2011)
	Oral motor skills – orbicular muscles	Guy and Neve (2005)
	Extensive vocabulary	Pineda and Pérez (2011)
	Enhanced pronunciation of phonemes	Pineda and Pérez (2011)
	Improved psycholinguistic skills: - Comprehension - Auditory association	Votoria-Gallastegi (2005(a)
	Communicate Feelings and thoughts through music	Bell (2008)

Table 1.4. Communicative elements influenced and developed by musical engagement in those with DS compiled from the review by Gemma et al. (Gemma, Pablo et al., 2020).

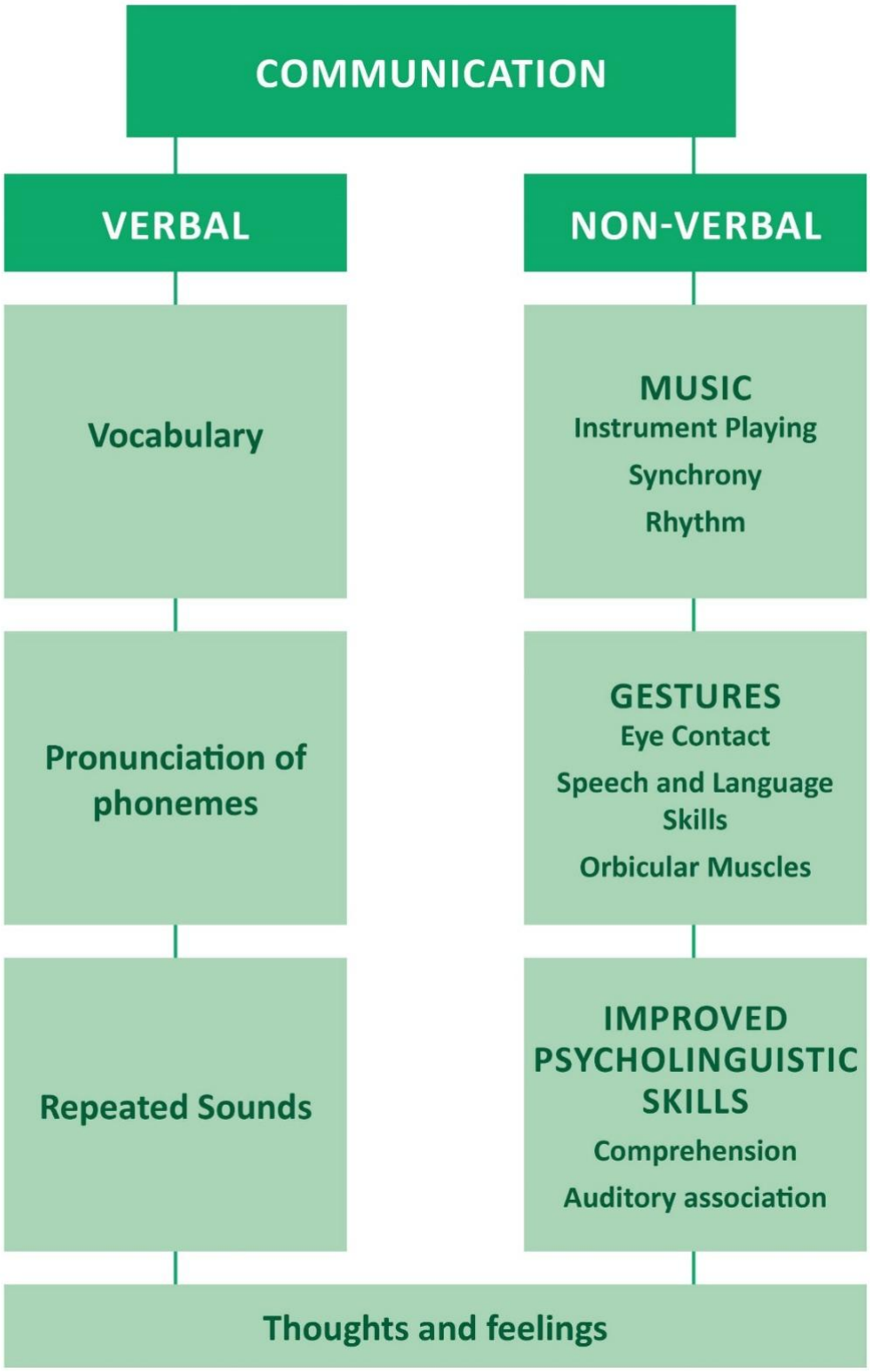


Table 1.5. Both Verbal and Non-Verbal communication are affected by music compiled from the review by Gemma et al. (Gemma, Pablo et al., 2020).

(c) Social and Emotional Skills

Improvements in social-emotional development have been noted across the studies described by Gemma et al., accounting for 37% of the articles studied. Both Polo-Dowmat (2002) and Guy and Neve (2005) emphasised the importance of musical engagement in promoting positive self-image and societal engagement (Polo-Dowmat, 2002; Guy & Neve, 2005). Guy and Neve noted the positive impact music experience has on confidence and how it increases productivity (Guy & Neve, 2005). In a study of thirty children that aimed to evaluate the effect of music on the anxiety of children attending a dentist, Gomez-Scarpetta et al. found that music therapy sessions relieved stress and anxiety (Gomez-Scarpetta et al., 2012).

Music engagement has also impacted trust and calmness, allowing those with disabilities to engage with their peers (Morgante, 2014; Camps et al., 2017). Arguedas-Quesada, citing Lagos (2004), noted how the participants' engagement with music helped them to relax, regulate emotions, and participate more fully in life activities (Arguedas-Quesada, 2015; Gemma, Pabolo et al., 2020). Mizumo and Sakumo found that playing a musical instrument assisted in raising the self-confidence of performers with DS (Mizumo & Sakumo, 2013). Pineda and Pérez conducted a descriptive study on the impact of music therapy sessions on 18 children with DS from a particular nursery in La Habana. The aim was to acquire responses from parents and teachers on the effectiveness of these sessions on the children. The teachers noted a 100% improvement in socialisation and learning, while 77.7% of parents noted improvements in these areas. Pineda and Pérez concluded that the early implementation of music therapy sessions in this cohort was recommendable, and more knowledge about music therapy is required for parents and teachers (Pineda & Pérez, 2011). This was also echoed in a later literature review by Theodorou and Drigas, who examined the use of music technology in therapeutic sessions with children with ID (Theordorou & Drigas, 2017). They found that technology has a vital role in music therapy sessions and helps to develop life skills in these children, but more knowledge around its potential usage needs to be researched further. A summary of the social and emotional areas impacted by music and the authors may be found in Table 1.6.

ELEMENT	AFFECTED AREA	AUTHOR
Social and Emotional Development	Relationships; Social Interaction	Polo-Dowmat (2002) Becker and Dusing (2010) Pineda and Pereze (2011) Pienaar (2012) Mizuno and Sakuma (2013) Morgante (2014) Theodorou and Drigas (2017)
	Self-Image/Self Confidence	Polo-Dowmat (2002) Guy and Neve (2005) Becker and Dusing (2010) Mizuno and Sakuma (2013)
	Engage more with Society	Guy and Neve (2005) Gutierrez, Ibanez, Prieto (2012) Arguedas-Quesada (2015) - citing (Lagos 2004)
	More productive	Guy and Neve (2005)
	Reduce Anxiety	Becker and Dusing (2010) Gomez-Scarpetta et al. (2012) Camps, Prina and Roderigues (2017)
	Improve Relaxation and Breathing	Gutierrez, Ibanez, Prieto (2012) Morgante (2014) Arguedas-Quesada (2015) – citing Lagos (2004) Camps, Prina and Roderigues (2017)
	Trust	Morgante (2014)
	Control Emotions	Gutierrez, Ibanez, Prieto (2012) Arguedas-Quesada (2015) – citing (Lagos 2004)
	Values	Pineda and Pereze (2011) and Theodorou and Drigas (2017)
	Empathy	Gutierrez, Ibanez, Prieto (2012) Arguedas-Quesada (2015) – citing (Lagos 2004)
	Understanding Rules	Pineda and Pereze (2011) Theodorou and Drigas (2017)

Table 1.6. The specific social and emotional skills positively impacted by music in those with DS compiled from the review by Gemma et al. (Gemma, Pablo et al., 2020).

(d) Cognitive Skills

A smaller number of studies have shown how music can influence cognitive behaviours. Table 1.7 shows the limited research into these most significant areas, with studies examining the impact of listening to familiar versus unfamiliar music and its effect on 'auditory-motor' coupling (Virji-Babul, Moiseev et al., 2012). Virji-Babul et al. aimed to explore brain activation in children with DS when listening to familiar and unfamiliar melodies played on the same instrument. They found that while familiar music was associated with 'auditory coupling', it did not activate brain areas involved in emotional processing in children with DS (Virji-Babul, Moiseev et al., 2012). De l'Etoile et al. examined the positive impact of a mother singing to a child with DS and how it affected attention (de l'Etoile, 2015). The researchers found that a mother singing to her baby for two minutes helped with attention, as reflected in the Infant Behaviour Rating Scales -Revised (IBRS-R) (de l'Etoile et al., 2015). Another study by Jiménez et al. also explored the effect of a cognitive intervention through music and its potential impact on visual and auditory STM. They found that a repeated rhythm motif on a drum and rainbow piano stimulated specific brain areas that had poor functioning in this cohort, including the HPC, the cerebellum, and the prefrontal lobe (Jiménez et al., 2011). In a preliminary study, researchers felt that exploring the role of neuroplasticity in music intervention studies could further enhance STM for visuospatial content (Jiménez et al., 2011)

ELEMENT	METHOD	AFFECTED AREA	AUTHOR
Cognitive Development	Listening to Familiar Music	Stimulated the left primary auditory region; left superior temporal sulcus and the right pre-motor region.	Virji-Babul, Moiseev et al. (2012)
	Mothers' directed singing for two minutes	Maintaining attention evaluated in accordance with the Infant Behaviour Rating Scales-Revised (IBRS-R)	de l'Etoile (2015)
	Intervention to stimulate visual and auditory short-term memory. Based on the repetition of short rhythmic stimuli with a drum and rainbow piano	Stimulated the hippocampus, the prefrontal lobe and the cerebellum.	Jiménez et al. (2011)

Table 1.7. The cognitive methods used and affected areas in those with DS, as found in the review by Gemma et al. (Gemma, Pablo et al., 2020).

(e) More recent Studies

Following Gemma et al.'s detailed review, research was carried out to see if any further studies had been published since the systematic review in 2020. As seen in Table 1.8, just two studies examined the impact of music engagement on children specifically with DS, while another included a DS cohort generally with other developmental profiles.

Title	Author	Date	Source
A Program for Developing Some Motor Skills for Down Syndrome Children Using Music	Huda M. Mazeed	2022	International Journal of Early Childhood https://doi.org/10.1007/s13158-022-00338-
Survey of the Home Music Environment of Various Developmental Profiles	Selena Steinberg, Carolyn M. Shivers, Talia Liu, Laura K Cirelli, Miriam D. Lense.	2021	Journal Applied Developmental Psychology https://doi.org/10.1016/j.appdev.2021.101296.Epub 2021 Ju l6
Locomotor and music: Influence on motor development in children with Down syndrome	Maria Ceclia, Nuala Izquierdo, Alejandro Jarrin.	2021	Revista Arbitrada Interdisciplinary Koinonía ISSN e (2542), 3088, Vol. 6, No Extra 2, 2021 (Issue dedicated to Especial Educacíon), 649–668.

Table 1.8. Studies that have been carried out since Gemma et al.'s review (Gemma, Pablo et al., 2020).

While only two examined the effect of music exclusively on children with DS, the other included a survey of a music environment that can affect children with various developmental profiles, including children with DS. Steinberg et al. wanted to see if a home music environment impacted children, both TD children and those with atypical developmental trajectories. Using an online questionnaire, the research team surveyed 340 families with children between six months and five years. They found that promoting music in a child's home environment and encouraging parent-led therapy through music enhances parent-child relationships (Steinberg, Shivers et al., 2021). While the study only included 25 participants with DS, these children showed significantly more engagement with music than other atypical cohorts.

The other two articles focused on strengthening motor skills in children with DS. The first was a bibliographic study centred on the impact of locomotor activities and music on this cohort. The study found that research projects that included music activities enhanced the motor skills of children with DS, but early intervention also significantly optimised the benefits. Most recently, a study explored the impact of a rhythmic programme on selected motor skills in children with DS (Mazeed, 2022). The study sought to discover the appropriate motor skills for a child with DS while also exploring what musical rhythms could be used to help develop these skills (Mazeed, 2022). The researcher wanted to see how effective melody could be in supporting motor concepts and subsequently create a motor skills scale for those with DS. The study randomly selected twenty children from a particular school in Giza. They were then divided into equal groups, one control and one experimental. The observation card scale was used in the pre-test for both groups and then the music programme was applied only to the experimental group.

Several musical activities were devised to assess their impact on the motor skills of the DS cohort. The programme involved 20 sessions that included an activity, such as a 'listen and imitate activity', a ball activity, and a train movement activity. In all activities, the experimental group showed dramatic improvements in all gross motor activities, including running, walking, jumping, and throwing. Therefore, the need for immediate interventions with the knowledge of the importance of rhythm, melody, and kinetics in the motor development of children with DS is vital (Mazeed, 2022).

There is a severe void in the number of substantiated studies examining the impact of music on people with DS, particularly children. As evidenced above, the methodologies and approaches vary considerably across many conditions. While remaining an underexplored field of research, much more investigation has been carried out on the impact of music on the TD population and other cohorts. Inspired by the existing studies, the current research project was created to address this void. Its elemental components are detailed in the following section.

1.13 Exploring MINdDS: Music as an Intervention in the Neurological development of children with Down Syndrome.

The current research project was created in response to the lack of consistent enquiry on the potential role music can have for children with DS. Entitled **M**usic as an **I**ntervention in the **N**eurological **d**evelopment of children with **D**own **S**yndrome (MINdDS) for convenience purposes, MINdDS was designed by the author through active research in collaboration with Music4Children, a music programme that promotes inclusion, accessibility, and fun. Founded by the author in 2011, Music4Children (www.music4children.ie) has worked with children of all abilities and created music programmes for several contrasting settings. Reflecting on this work, the content for MINdDS was carefully designed and formulated using music education techniques with additional new and innovative approaches to teaching music, including neuroscience, early childhood education, cognition, and therapeutic forms of engagement.

The MINdDS project not only contains a diversity of musical content but is also navigating unchartered territory as it straddles the intersection between music education and music therapy to accommodate all learners.

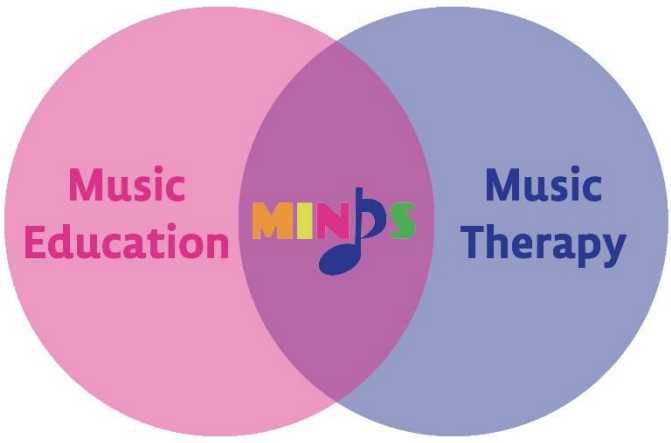


Figure 1.30. A visual representation showing the overlap between the fields of music education and music therapy in the MINdDS project.

Music education may be defined as a field of practice that teaches children the fundamental components of music. Specifically, "it encompasses music-generative

activities such as composing and improvising, performing opportunities, and learning about and listening to music" (Murphy & Fautley, 2015). However, adhering to music education *only* may limit the accessibility and potential impacts of the intervention on this cohort. It became evident that there was a need to look concurrently at the field of music therapy as some of the therapeutic approaches and ideals coincided with the perspectives in the project. Music therapy…

> engages people in a variety of musical experiences in order to restore, maintain or improve functioning and quality of life. It can focus on cognitive, physical, psychological, emotional and social needs and uses evidence-based interventions to meet clinical goals in these domains (Ahessy, 2014).

With similar underpinning ideals, MINdDS, encourages both active and receptive interventive methods found in music therapy practices. In addition, music therapy acknowledges the importance of nonverbal communication through music and how this can impact learning and personal development, an important consideration with this cohort and applied by the MINdDS approach (Gilroy & Lee, 1995; Jimmy Petruzzi, 2023

However, the preference for music therapy when engaging children with DS in music has become evident throughout the literature review (Gemma, Pablo et al., 2020). It has also exposed the rich educative/creative space between music therapy and music education, wherein this project lies. However, there is little acknowledgement of this 'in-between space' within the literature (Raglio et al., 2011). The language used around this work exacerbates this divide, with 'interventions' being the preferred term when teaching music to children with disabilities and 'music training programmes' when teaching TD children (Miendlarzewska & Trost, 2014; Shen, Lin et al., 2019; Gemma, Pablo et al., 2020). What has been emphasised in the literature review is that the content in these music training programmes is often similar to the content found in the interventions that are used with children with DS (Dumont, Syurina et al., 2017). Reflecting this, both 'training' and 'intervention' are used throughout this thesis, with the word 'training' referring to the music programme content leading to knowledge acquisition, while the term 'intervention' refers to the action taken to help improve outcomes for children through music.

While in no way claiming the clinical qualification of a music therapist or their significant role in clinical practice, this study lies comfortably between the fields of music education and music therapy. Therefore, MINdDS is a music programme that delivers the fundamental components of music through active and receptive methods to help enhance the quality of life of all children by engaging cognition, physiological movement, communication and social and emotional skills. How this new hybrid field was shaped will form the focus of the sections that follow, which will help explain the approach taken and why.

1.13.1 Neuromyths, Neuromania and Neuroskeptics: The Role of Neuroscience in MINdDS

The role of neuroscience in creating the intervention was considered not only from the many measures that accompanied the study but also in understanding the neuroscience of learning. While neuroscientific research has gained ground in other disciplinary settings, there are still many challenges in creative environments to accept the potential role and contribution this field of study could have to our increased understanding of music and its impact on the brain.

There has been much controversy around the emphasis placed on neuroscience in learning, particularly in relation to early years education and its impact on creativity. Susan Young, a well-respected researcher and practitioner in early years education, has done pioneering work in the music education sector. Young understandably challenges the role of neuroscience in creative learning, as they believe the neurocentral approach to be very detached from

> social, material and cultural context, represent[ing] a highly reductionist, pure-science point of view. The child has become merely a biological entity, an organ. From a social constructionist point of view, a focus on the 'brain' results in an extremely impoverished construction of children in which mechanistic brain functions predominate and the subjectivity of the child the mind of the child as thoughtful, emotional, social, and cultural, in a living, breathing body, is completely erased (Young, 2018).

This is supported by John Bruer (2002) in their 'Myth of the First Three Years' and by the Organisation for Economic Co-Operations and Development, which defined the term 'Neuromyth' (OECD, 2007). Many other terms have been coined in response to the increased interest in neuroscience, such as 'Neurocritic', 'Neuroskeptic' and

‘Neurobonkers’. Young warns of the ‘seductive allure’ of science and cites Goswami as

> the worry is that in the field of early childhood music education, unsubstantiated claims about how children’s brains can be changed through musical input may lead to the implementation of practices that seem appealing but will have little benefit whatsoever (Goswami, 2006).

The ‘neuro’ prefix has been applied to several subjects and contexts in what Legrenzi and Umilta have entitled Neuromania (Legrenzi & Umilta, 2011; Tallis, 2011; Wastell & White, 2017). The authors assert that “science is not an anthropomorphic being. It does not ‘tell’ anything. Scientific data has no meaning until one interprets it. Such interpretations are inevitably packed with qualitative judgements” (Wastell &White, 2017). They continue that ‘brains pack a punch’ and that the brain is charismatic and appeals to people (Wastell & White, 2017). Such information can, of course, be used in an isolated way, creating a disconnect between the person and the data. However, I believe there is a place for neuroscientific research in the holistic view of teaching and learning in the arts and, hence, creating a suitable music education programme. There seems to be the ‘either-or’ stance – rather than excluding one over the other, neuro data should be included in an overall approach that considers the child/participant holistically. The potential benefits of such an approach for motor skills, auditory processing, and communication have been previously noted. Therefore, the neuroscience of our brains should play a role in developing these abilities and skills.

This interrelationship inspired the neuro-causal model created in the introduction to this thesis. It is re-presented here to re-establish the author's viewpoint and provide the context for creating the project. This emphasises the importance of neurological, biological, physical, and environmental factors, supporting and enhancing creativity rather than stifling it.

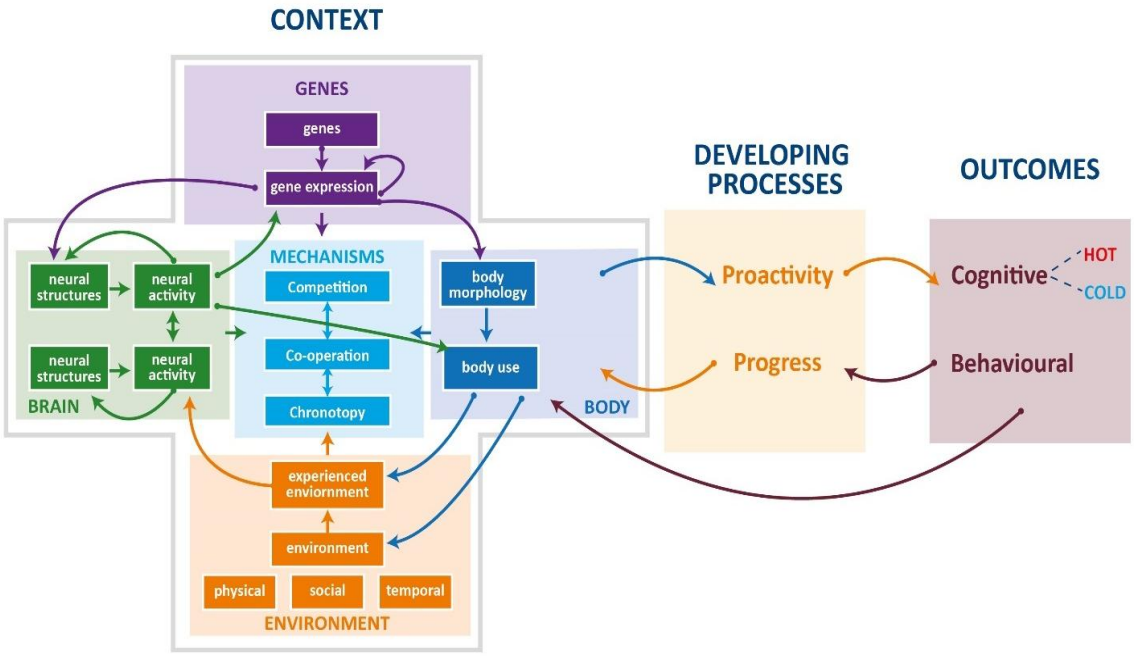


Figure 1.31. A restatement of the neuro-causal model for considering atypical development was formed from the neuroconstructive model of Westermann, Mareschal, Cebula, and Moore (Cebula, 2010; Westermann, 2010).

1.13.2 The Context: Neural Education as a Pedagogical Model

The importance of brain morphology and its impact on abilities to receive, process and store information has been previously detailed. Educators and pedagogues encounter challenges when differences occur in what is categorised as 'normal' or 'typical' brain structures, as these can challenge existing teaching methodologies. The delivery of the MINdDS intervention has also been influenced by a model of neural education devised by Dr Kieran O'Mahony and Richard Carr. In their brain-centric design, they emphasise how important it is to teach to people's brain types to optimise their performance. Optimising human performance is central while creating a love of learning is at the core of the approach. Carr and O'Mahony have recognised the difference between learners and the need to consider neuroscience when teaching children. Inspired by a publication, *The Orchid and the Dandelion* by Thomas Boyce, their teaching method helps accommodate the most sensitive learners. It enables them to thrive, most importantly, alongside their peers. This method is based on cognitive neuroscience; therefore, activating specific brain areas has enhanced the learning process for many children.

The acknowledgement of physical and environmental factors outside brain morphology must also be considered, as seen in Figure 1.31. This is supported by Sinnott, who believed that:

> [t]he external configuration is usually relatively simple, but there is packed into the interior of an organism an amazing complexity of structures which have long been the delight of anatomists. The specific form of a plant or animal is determined not only by the genes in the organism and the cytoplasmic activities that these direct but by the interaction between genetic constitution and the environment. A given gene does not control a specific trait but a specific reaction to a specific environment. (Sinnott, 1963).

Citing John Medina, a molecular biologist, Carr and O'Mahony similarly believe that:

> [i]f you wanted to create an education environment that was directly opposed to what the brain was good at doing, you probably would design something like a classroom. If you wanted to create a business environment that was directly opposed to what the brain was good at doing, you probably would design something like a cubicle. And if you wanted to change things, you might have to tear down both and start over. (O'Mahony & Carr, 2019).

This brain-centric approach is based on creating relevant content to 'ME HERE NOW' (O'Mahony & Carr, 2019). The neural education model puts children at the centre of this approach. For a child to effectively learn, all brain parts must be engaged. This is encouraged by first activating the occipital lobe using adequate visual information. The parietal and temporal lobes are activated by providing opportunities to use small motor skills and vocalising. Asking the learner to teach the concept back to you while showing what the wrong answers could be is also inherent in the design.

It has been proven that children learn best when they are in their PFC, allowing access to their EF, which is needed to optimise learning (O'Mahony & Carr, 2019). We have already seen that children with DS have a normal amygdala and, therefore, are susceptible to the same 'flight, fright and freeze' reactions as TD cohorts. The amygdala is named after the Greek word for almond and is what the authors call the world's gatekeeper (O'Mahony & Carr, 2019). The amygdala is part of the limbic system. It is located very close to the HPC, which, as we have already seen, is responsible for learning and memory and is significantly reduced in those with DS. When an amygdala hijack occurs, the child or student cannot learn. Physiologically, they cannot access their PFC and cannot problem-solve or acquire knowledge. The HPC is where neurogenesis and plasticity can occur; therefore,

children with DS could benefit hugely if an appropriate and positive intervention is created.

While limiting cognitive load, cognitive rehearsal is essential within this pedagogical approach, strengthening brain connections. This capitalises on practice creating myelination and strengthening brain networks, hence connections between brain areas. Therefore, this intervention is based on the idea that 'practice makes permanent' and includes much repetition, which is integral in the learning process for children with DS (O'Mahony & Carr).

The prevalence of reducing cortisol and increasing oxytocin in previous music studies has been emphasised. Therefore, this intervention has been aimed at reducing the stress in learning contexts while heightening oxytocin levels. Stress has been linked to increases in cortisol found in the hypothalamus and the amygdala, and it causes a loss of synaptic connections between neurons that can impact the PFC. Oxytocin is found in reproduction, social learning, and affiliative behaviours (Harvey, 2020). It also assists in the development of many cognitive functions and motor skills. It is believed that music, dance, and social bonding move people into their PFC as "oxytocin's effects on social recognitions, learning and memory are associated with activity in the hippocampus, amygdala, nucleus accumbens, and prefrontal cortex." (Fergusson et al., 2002; Hurlemann et al., 2010; Mitre et al., 2016; Greenwich et al., 2018; Harvey, 2020). Of particular interest is the impact of music on the plasticity of the HPC (Herdener et al., 2010) and how it might be related to oxytocin levels. It is believed that oxytocin can increase long-term potentiation (LTP) levels and hence increase learning. It is thought that oxytocin also protects the HPC from stress. Harvey believes that knowing music's neurogenic impact on the HPC and its implication on oxytocin should be coupled with dance and movement, as movement is entrained through rhythmic synchrony.

Music affects many aspects of our lives, both extrinsically and intrinsically. Providing enjoyment can also significantly impact the development of perception, cognition, and motor skills (Harvey, 2020). Music also enhances our ability to engage with people socially and, as it is associated with the limbic system, is connected to the reward system, motivation, learning and memory (Zatorre & Salimpoor, 2013; Koelsch, 2018; Harvey, 2020). The impact of listening to music on biological responses

in clinical and nonclinical settings has recently been reviewed. It has revealed that the predominant biomarker affected was the stress hormone cortisol, with half of the clinical studies demonstrating a stress-reducing effect of music listening in clinical settings (Finn & Fancourt, 2018). Many of the other biomarkers analysed are also part of biological stress pathways, which suggests that the primary way by which music listening affects us biologically is via modulations of a stress response. Studies have demonstrated that salivary levels of the hormone oxytocin, linked to social bonding, increased in response to music listening (Harvey, 2020). These results suggest that listening to music affects us biologically by alleviating anxiety/stress and promoting social bonding.

However, the impact of music interventions on sociability has not been investigated nor explored in relation to children with DS who experience limitations in both attention and sociability. One of the fundamental vehicles for encouraging and facilitating this approach is through the medium of play and games and enjoyment as a vital framework in conjunction with this model of neural education. Neural Education believes that games and play help to create optimum learning environments within children as they help to release oxytocin and decrease cortisol levels, bringing children into their PFC and enhancing learning opportunities.

1.13.3 Play, Games, Dreams and Proximal Zones: The Importance of Play in a Learning Environment

Inspired by the work of theoreticians Piaget and Vygotsky, this section explores the importance of play in creating the optimum learning environment for children. After their publication *Play, Dreams and Imitation in Childhood*, Jean Piaget believed that a child's learning was at its highest potential when it was done through play. The psychologist focused specifically on the concepts of assimilation and accommodation. The first "incorporates events, objects, or situations into existing ways of thinking, which constitute organised mental structures," whereas, in accommodation, "the existing mental structures reorganise to incorporate new aspects of the external environment (Nicolopoulou, 1993)." However, play prioritises assimilation, incorporating events and objects with existing mental structures. Piaget's approach details how a child's play develops in their first seven years of life.

While the importance of play has a detailed history, its significant role in education and how it can enhance children's learning is still misunderstood in some contexts.

In English, games and play are defined as contrasting entities, as opposed to languages such as German, Spanish, and French, where they are seen as synonymous. In the English language, games and play are understood as related yet separate entities, and therefore, the importance of play as a legitimate educational method has often been undervalued. This was found in a study by Koutsoupidou, who asked teachers in early years settings to evaluate the importance of play (Koutsoupidou, 2020). At the same time, the significance of play in childhood development is receiving increasing attention in developmental approaches, particularly in anthropology, sociology, education, and psychology. However, in this study, play is still associated with entertainment. Only 20% of early practitioners believe play is essential in learning, whereas 40% believe it is imperative in entertainment (Koutsoupidou, 2020).

Interestingly, games are often defined differently as Schell believes “a game is a problem-solving activity, approached with a playful attitude” (Schell, 2008). The Music4Children programme believes that there is a substantial overlap between the two and that one depends on the other (Figure 1.32).

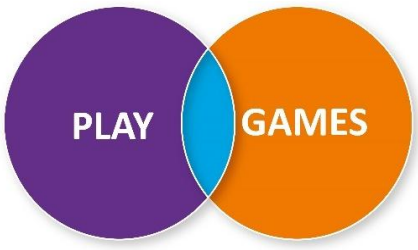


Figure 1.32. Music4Children believes that there is a distinct overlap between the play and games and that one is dependent on the other.

This is supported by Walz, who states that play is a game's foundation and that they depend on each other. (Walz, 2010). Both can enhance and create several learning opportunities.

Larson highlighted the importance of a third element of make-believe. He thinks play has two distinct yet interconnected layers, creating a spatial dyad (Larson, 2014). The first layer is the environment in which the play takes place, both inside/outside. The second layer puts the play in a meaningful context. He states that "it can be argued the assigned context is analogous to mental augmentation or expansion of the ongoing activity." (Larson, 2014). Larsen gives the example of a child climbing. There is a distinct difference between a child climbing and a child playing climbing; the latter requires mental augmentation to enhance its meaning and significance and encourages further connections and myelination in the brain (Larson, 2014). Larson's ideas are embedded in the theories of Vygotsky, who believed that genuine play begins at three years of age with pretend play. They believe that all play should be a continuous social construct. Hence, it "typically involves more than a single child; and the themes, stories, or roles that play episodes express children's understanding and appropriation of the sociocultural materials of their society" (Nico, 1993). Larson also highlights the importance of independent play as a social interaction with a cultural framework. As stated by Vygotsky, "what passes unnoticed by the child in real life becomes a rule of behaviour in play." (Vygotsky, 1933/1967). They also believe play to be vital in perception and language – satisfying needs to what is absent (Larson, 2014). Larson gives an insightful meaning to the transition of thought that occurs and cites Vygotsky. They use the analogy of a horse – that, at first, a child cannot think about a horse without seeing the image. However, over time, they can depict a horse using a stick; "the stick becomes the pivot for detaching the meaning of 'horse' from the real horse" (Larson, 2014).

Pretend play, or what Piaget called symbolic play, has formed an integral part of their framework. Piaget identified differences between contrasting kinds of play, including practice play, symbolic play, and play without rules, as the successive forms of play that occur in four development stages: sensorimotor, preoperational, concrete, and formal operations, as seen in Figure 1.33.

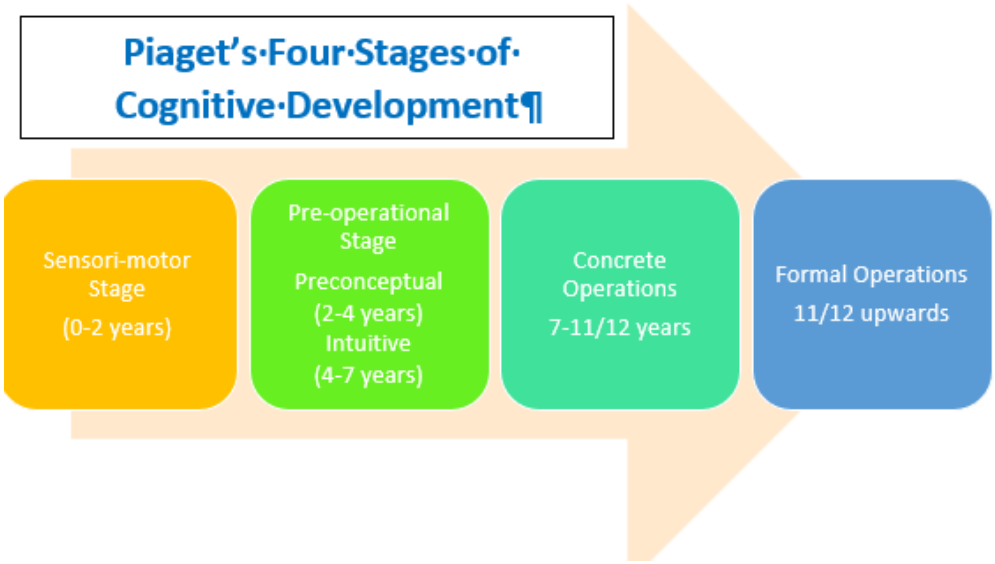


Figure 1.33. Adapted from The Four Stages of Cognitive Development proposed by Piaget as seen in <https://www.theneurotypical.com/piagets-four-stages.html> accessed May 2021.

Symbolic play occurs in the second year of childhood, is associated with pretend play, and coincides with language development. This overlaps with Piaget's preoperational stage of development. While symbolic play, according to Piaget, declines at age four, symbolic play forms a considerable part of this intervention. At the age of 4, there is increased questioning, referred to as 'the birth of primitive reasoning'.

Piaget also believed play was for enjoyment, as the citation taken from their *Play, Dreams and Imitation in Children* (1962) depicts

> unlike objective thought, which seeks to adapt itself to the requirements of external reality, imaginative (make-believe) play is a symbolic transposition which subjects things to the child's activity without rules or limitations. It is, therefore, almost pure assimilation, i.e., thought polarized by a preoccupation with individual satisfaction (Piaget, 1962; Larson, 2014).

Therefore, play, games, and make-believe are inherent in increasing the learning environment for children (Figure 1.34).



Figure 1.34. Music4Children believes in the inherent link between make-believe, games, and play that helps to scaffold and optimise learning.

The MINdDS music intervention included material that would promote and foster different forms of play to assist all learners to engage with the material. Therefore, games, play, and make-believe were intrinsic to the programme to encourage contrasting levels of engagement. However, the 'role of the adult' in play was just as crucial in the adopted methodologies. Like Vygotsky's theory, the programme believes that play contributes to cognitive development rather than reflecting it. Vygotsky believes that "instruction is good only when it proceeds ahead of development. It then awakens and rouses to life those functions which are in a stage of maturing which lie in the zone of proximal development." (Vygotsky, 1967). The Zone of Proximal Development (ZPD) is crucial in the elemental design of the intervention. Scaffolding learning through the support of the interventionists and parents is vital to the programme's success. Seeing the potential of children achieving beyond what they can do on their own is invaluable for all – the 'productive struggle' with additional technology support, as seen in Figure 1.35.

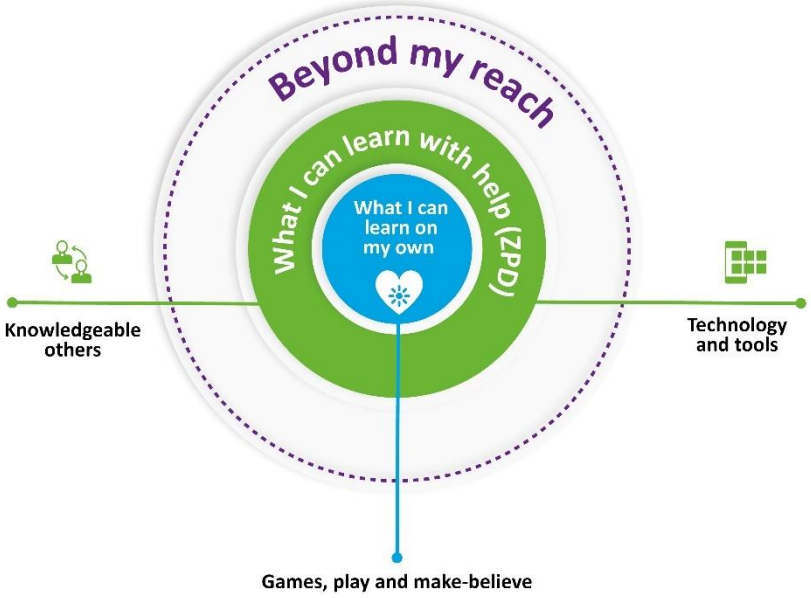


Figure 1.35. Vygotsky's Zone of Proximal Development was modified by McCleod (2018) and Lim et al. (2018). These were then collectively adapted for the MINdDS intervention, which includes the elements of games, play, and make-believe.

In conclusion, the components of game, play, and make-believe, along with the concepts promoted by Piaget and Vygotsky, have been inherent in devising the programme, see Figure 1.36.



Figure 1.36. Piaget's Symbolic Play stage and Vygotsky's Zone of Proximal Development, support the importance of play, games, and make-believe in the MINdDS intervention.

1.13.4 Let the Music Play: Embodying Musical Play within a Social, Physical and Temporal Environment

Many pioneers of musical play were imbedded in musical teachings throughout the early-mid twentieth century. Kodály, Dalcroze, and Orff promoted and developed more creative teaching methodologies to increase the accessibility, practice, and learning of music in educational settings. Each promoted the importance of musical play but specialised in particular approaches, as seen in Figure 1.37.

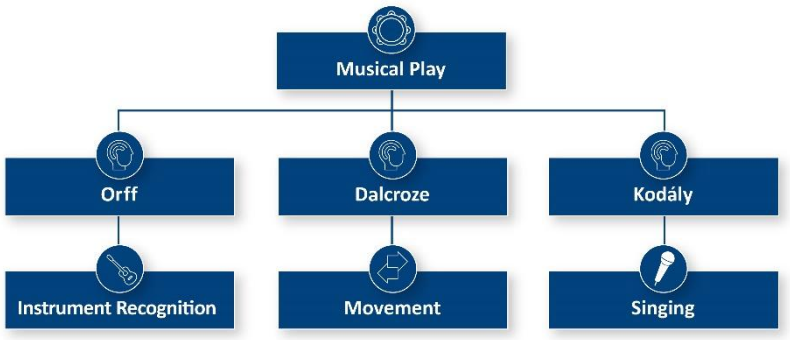


Figure 1.37. The teaching methodologies as promoted by Orff, Dalcroze, and Kodály.

Émile Jaques-Dalcroze (1865–1950) developed methods for students to engage physically with music concepts to enhance their understanding. Dalcroze intended to teach musicality in an innovative way that would bring these musical concepts to life. They divided their methods into eurythmics, which used rhythm and movement to help students better understand the concepts of solfège, the study of pitch, and improvisation.

However, Dalcroze is renowned primarily for eurythmics and their work with rhythm. (Hall, 1920). It is believed that musical rhythms help to orient attentional processes in time, which implies a benefit for the preparation and control of motor actions and, therefore, the enhancement of the temporal processing of information (Miendlarzewska & Trost, 2014). In addition, Miendlarzewska suggests “that musical activities that imply perception and production of rhythms train attentional

processes which benefit other cognitive functions" (Miendlarzewska & Trost, 2014). An explicit relationship between rhythm and movement may be found in the locomotor rhythms they used to describe rhythmic note values. A giant step denotes a two-beat minim, a crotchet as a march and tip toe for the quavers. The importance of physically relating the musical rhythms of the Music4Children rhythm family to movement was fundamental in the music intervention, which similarly was to heighten the children's understanding of the concepts.

Kodály (1882–1967) believed in teaching children music that was intrinsic to their culture and relatable. They developed solfège hand signs, which have been used in various contexts to help teach children music and song (Burns, 2020). Kodály believed the voice to be the most accessible instrument and, therefore, devised hand signs to achieve more accurate pitching. Within the intervention, this approach was modified to include children's names and more physically accessible signs that resembled the visual characteristics of musical notes.

Carl Orff (1895–1982) believed every child could learn music if presented in an accessible, relatable, and fun manner. The American adaptation of Orff's work uses four stages of learning: imitation, improvisation, exploration, and literacy (Beegle & Bond, 2016). Orff's method also includes narrative through movement and drama that reflects a child's world of play.

While these are just some of the methodologies steeped in the historical context of musical play, using play as a medium to teach music is still an under-explored methodology in practice. Therefore, musical play is at the core of the intervention to illustrate how minor modifications in teaching practice can increase the accessibility of music to children of all abilities.

1.13.5 Losing the Plot: The Potential Role of Narrative in the Socialisation of Musical Play

Storytelling is a fundamental component of children's lives in educational settings, at home or through screen time. The benefits of storytelling and narration have gained importance in educational settings to enhance the learning experience of diverse curricular subjects (Irwin et al., 2012). It now takes on and is presented in many varied and contrasting forms. A recent study by Nicolopoulou in 2015 showed that narration

and play promoted literacy and social skills in children of low-income families (Nicolopoulou, 2015). Entitled 'Using a narrative- and play-based activity to promote low-income preschoolers', the author researched the potential of storytelling and story-acting practice (STSA) to enhance a child's readiness for school. This included their oral language skills, literacy, and social competencies. There were 149 families of 3–4-year-olds in the six experimental classrooms and seven control groups. It was found that those children exposed to the STSA curriculum displayed a readiness to school in promoting learning and literacy in these children.

Learning music is often associated with the perfected performance of an instrument (Sarazin, 2016). Even though the role of music in early childhood settings has been mentioned previously, music is still seen as a specialised language that is only accessible to those who have received professional training (Young, 2016). However, creating a narrative tool or a medium can help break down these barriers, increasing the accessibility of music to, not only the children, but also the practitioners or facilitators. There is now a wealth of picture books with music that can create an excellent, scaffolded environment to help engage children's musical imagination. In *Stories and Narratives as Agencies of Change in Music Education*, Westerlung highlights that "the power of a story to capture the imagination depends not just on *what* we narrate but, to a degree, on *how* we narrate in communities" (Westerlung, 2020).

Through this intervention, narrative forms an important component, explicitly using puppets. The 'Amazing Benefits of Playing with Puppets in Early Childhood' was acknowledged by Mcilroy, who believes that it supports the verbal expression of children and provides a medium for children to express emotions while also assisting with memory (Mcilroy, 2020). In addition, "puppets lend themselves to being the character in your stories and allow your child to join in with the action" (Mcilroy, 2020). The role of the rhythm family formed a critical element in each lesson plan. Narratives around these were to enhance the children's understanding of the music. The rhythm family was also embedded in the animated clocks created with the children, as seen in Appendix E.

1.13.6 What is on Our MINdDS in Summary

The previous sections have mapped the pedagogical reasoning and rationale for devising the intervention that has led to the elemental components of music learning, a summary of which may be seen in Figure 1.38.

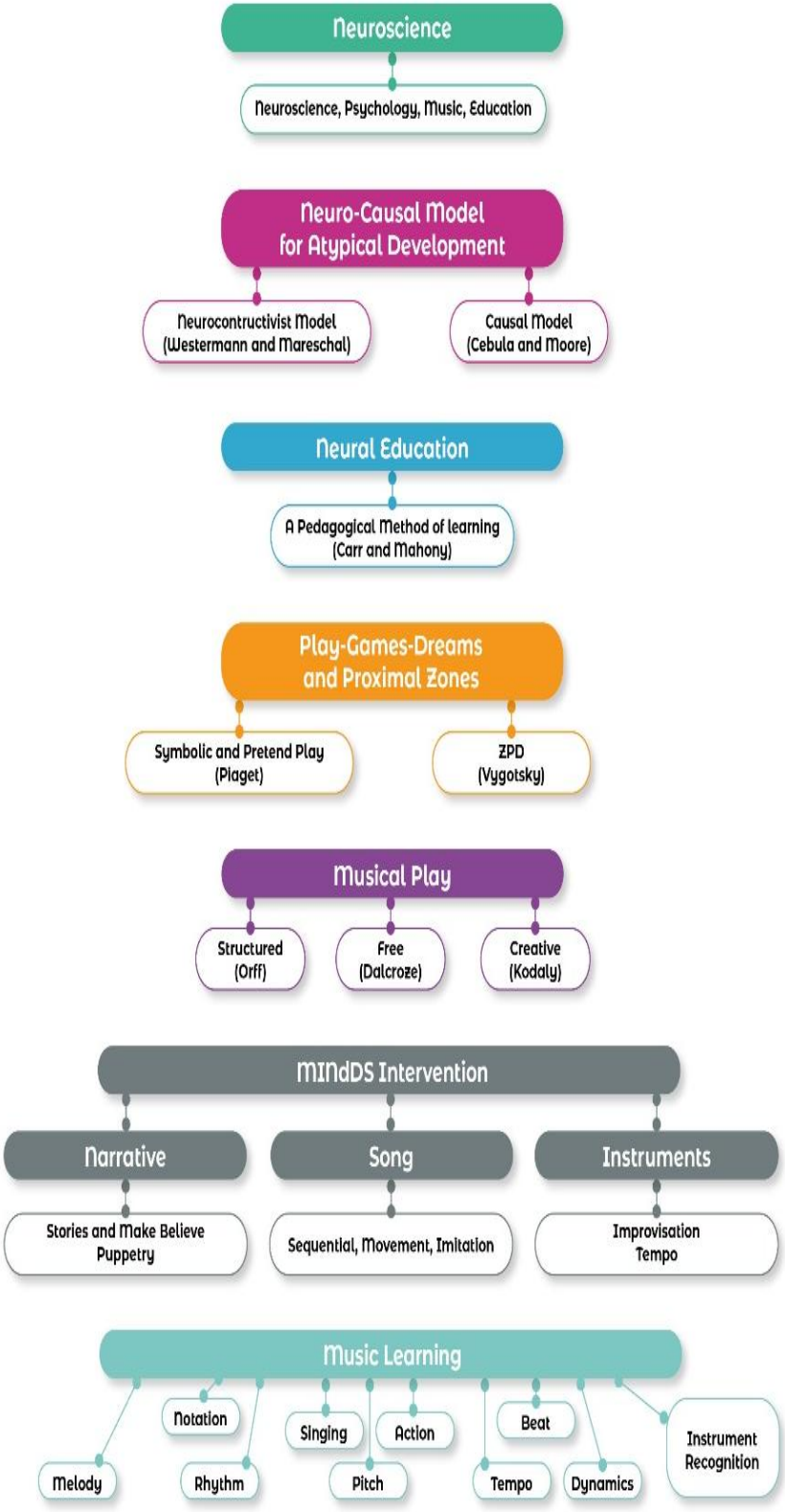


Figure 1.38. A detailed overview of the pedagogical methodologies embedded in the MINdDS intervention.

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These contextual components are intrinsically based on the neuro-causal model that has formed the centre of the intervention that frames this research project. Each intervention strategy is interlinked with the neurobiological and environmental contexts, a summary of which may be seen in Figure 1.39. The aims and processes involved in the project are detailed in the sections that follow.



Figure 1.39. The intervention strategies are related to the neurobiological and environmental contexts.

1.14 Aim of this Study

The study aims to test potential improvements in temporal perception, visual memory and alterations to cortisol and oxytocin after a music intervention with TD children compared to children with DS. Music can potentially improve behavioural outcomes in children with ID. However, the specific benefits of music on cognitive abilities need to be better understood. The only way to improve these interventive techniques is to know how they function and the potential impact they may have, if any, in the daily lives of children.

Collaboration with Down Syndrome Cork (DSC) and Music4Children (with Dr. Eva McMullan) is ongoing to deliver online music programmes for TD children and children with DS based on pre-COVID experiences. The positive effects of music on children with DS have been noted locally by parents and teachers who have anecdotally reported increased attention and self-monitoring. In fact, *"*the amount of benefits these sessions have on auditory processing, language skills, speech, shared experience with parents, attention building, turn taking, literacy skills and most of all fun is astounding" (Alison Wilson – DSC). However, how cognitive and biological changes relate to the anecdotal parental-reported behavioural improvements in children with DS because of music is not fully understood. For example, it is unknown whether music improves early sensory processing or higher-order cognition in children with DS, promotes sociability/or alleviates anxiety through biological changes.

Therefore, the study aims to test these potential improvements in temporal perception, visual memory, cortisol and oxytocin after a devised music intervention. The intervention will be conducted with children with DS who are MA-matched with their TD peers. The study will look for potential confounding factors to ensure no changes were due to developmental alterations. To assess biological changes associated with stress and sociability in children, both cortisol, the hormone linked to stress, and oxytocin, the hormone linked to social bonding, will be measured in saliva and hair samples pre- and post-intervention.

To achieve these aims, the following questions will be addressed.

1. What is the recruitment profile of both the TD and DS cohorts that are engaged with the study?
2. What is the current availability of music tuition?
3. What has been the children's experience of the intervention?
4. What has been the impact on temporal perception as a result of the intervention?
5. What has been the impact of the intervention on short-term spatial memory?
6. Has the intervention impacted the quality of life of the participants, specifically physical functioning, emotional functioning, social functioning, and school/work functioning?
7. How did the music intervention impact cortisol and oxytocin concentrations in saliva and hair samples?

Methodologies

2.1 Ethical Approval

This study was initially approved on December 21st, 2019, by the Clinical Research Ethics Committee (CREC) at University College Cork. The initial submission included in-person music classes supported by music videos explicitly designed for children. However, due to COVID-19, the delivery of the music intervention had to be reviewed and subsequently moved online. The online approval from CREC was received in June 2021, and the Greenlight was obtained in July 2021 (Figure 2.1).

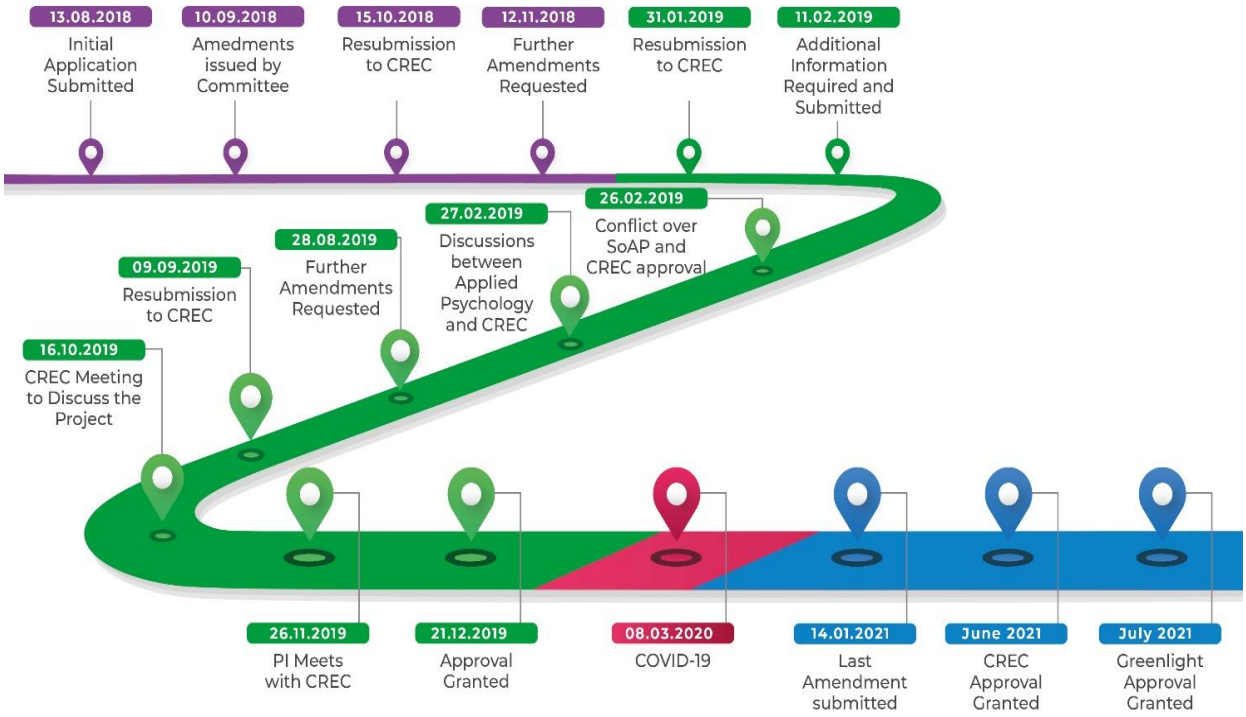


Figure 2.1. A detailed timeline showing the journey to ethical approval from the initial ethics application submitted on 13th August 2018 to the Greenlight approval awarded in July 2021.

2.2 Study Design

The study was designed to measure the effects of a music intervention on the cognitive abilities of children with DS compared to TD children (Mertler, 2011). A pre-test and post-test design was applied (Mertler, 2011). There were 55 children recruited at the start of the study: 33 TD children and 22 children with DS. Each child was given a number in their respective cohort, and using a Google random number generator, they were assigned to either a control group that received no music or a music intervention group that received music. For convenience, the study was called MINdDS, **M**usic as an **I**ntervention in the **N**eurological **d**evelopment of children with **D**own **S**yndrome. The breakdown of the respective groups may be seen in Figure 2.2.

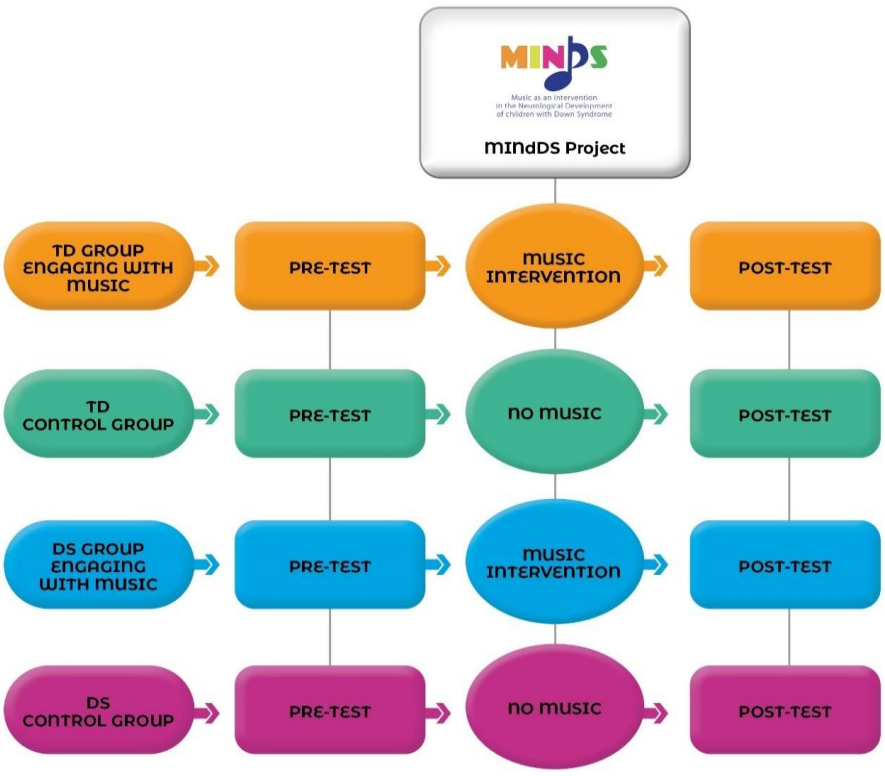


Figure 2.2. The experimental design adopted in MINdDS shows four distinct groups: two control groups (one TD, one DS) and two experimental groups (one TD, one DS).

2.3 Recruitment Process

Recruitment started in August 2021, with the project being advertised through UCC Outlook, the project's designated website https://www.ucc.ie/en/mindds, Down Syndrome Cork (DSC) and through the Templebreedy National school website

https://templebreedyns.ie. The advertisements specified that only TD children aged between 4 and 8 years could be involved in the study. Meanwhile, the age range for those with DS was 4 to 12 years. The children interested in engaging with the study could not participate in a daily music programme and had not been given an additional diagnosis. Full details of the inclusion and exclusion criteria may be seen in Table 2.1.

Inclusion Criteria	Exclusion Criteria
1. Children with Down Syndrome who are aged between 4-12 years. 2. Down Syndrome children that have not, to date, engaged with a daily music intervention programme. 3. Down Syndrome children who are engaged in the Music4Children programme. 4. Down Syndrome children who are not in receipt of any other formal music education outside the research context. 5. Typically developing children who are aged between 4 and 8 years. 6. Typically, developing children who are engaged in the Music4Children programme. 7. Typically developing children not engaged in the Music4Children programme or any formal music education programme outside the research context.	1. Children with Down Syndrome who are engaged in music education or music therapy sessions already. 2. Typically developing children engaged in a music programme outside of this study. 3. Typically developing children diagnosed with a learning difficulty before this study. 4. Down Syndrome children and typically developing children are outside this study's age profile.

Table 2.1. Inclusion and Exclusion Criteria for the MINdDS project.

Before the commencement of the project, an information evening for interested parents and guardians was held on MS Teams detailing what was included in the study. Those interested in meeting the inclusion and exclusion criteria emailed the lead investigator and were sent the relevant links to the information sheets, consent, and assent forms through Qualtrics, a survey tool approved by University College Cork (see Appendix A). The parents/guardians of the participants were invited to create a unique subject code on the assent form used for each questionnaire and

sample to ensure blinding. After completing the consent process, the parents were also required to answer two questionnaires, 'Why Choose Music?' (Paterson, 2008) and the Paediatric Quality of Life (PedsQL) (Varni et al., 2001) form before the intervention period.

Due to the limited number of recruited participants, the study was rolled out twice. The first wave took place between September 2021 and December 2021. Figure 2.3 shows the sequence of how each component of the control and music intervention groups was administered in wave one.

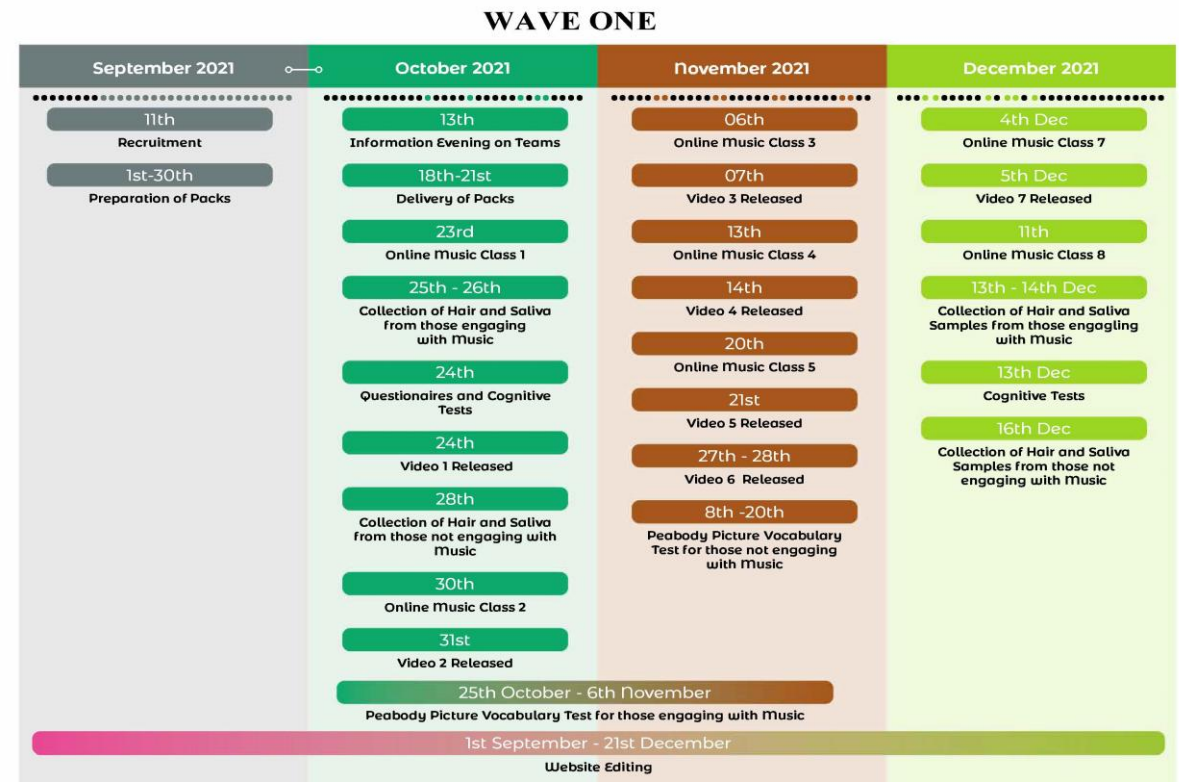


Figure 2.3. Timeline: interventions and tests and when they were administered in wave one.

In wave one, 36 participants were recruited: 22 in the TD group (M=12; F=10) and 14 in the group of children with DS (M=7; F=7). No one withdrew from the DS groups, and 5 withdrew from the TD group (M=4; F=1). This included one participant withdrawing from the control group (M=1) and four participants from the music intervention group (M=3; F=1).

Wave Two occurred between January 2022 and April 2022 (Figure 2.4).



Figure 2.4. Timeline: interventions and tests and when they were administered in wave two.

In wave two, 19 participants were recruited: 11 in the TD group (M=4; F=7) and 8 in the group of children with DS (M=6; F=2). Two TD participants withdrew from the music group (M=1; F=1), and two children with DS also withdrew from the music group (M=1; F=1).

Therefore, 55 participants were recruited in both waves (M=30; F=25), 33 TD (M=17; F=16), and 22 children with DS (M=13; F=9). While the initial wave was limited to the Cork region, the second wave was extended nationwide, as seen in Figure 2.5.



Figure 2.5. The demographic outreach of MINdDS in waves one and two.

Across both waves, there were 20 participants in the TD music group (M=11; F=9) and 13 (M=6; F=7) in the group that did not receive music. Seven participants left the study: 6 (M=4; F=2) from the music group and one from the control (M=1). This left 26 participants in the TD group (M=12; F=14) who completed the study. There were 22 children with DS in both waves (M=13; F=9); 9 (M=6; F=3) participants in the control group and 13 (M=7; F=6) in the group that engaged with music. Two (M=1; F=1) participants withdrew from the music intervention group.

Therefore, the total number of participants to complete the study was 46. There were 26 TD children (M=12; F=14) and 20 children with DS (M=12; F=8). The

groups were evenly matched, with 25 participants involved in the music group and 21 participants not participating in the musical intervention. A summary of the numbers involved in the study may be found in Table 2.2.

Wave	Total Number of Participants	Break Down of Total TD *n*	DS *n*	Music Intervention TD *n*	DS *n*	Control (No Music) TD *n*	DS *n*
Recruitment Wave One	36	22 M=12; F=10	14 M=7; F=7	13 M=8; F=5	7 M=3; F=4	9 M=5; F=4	7 M=4; F=3
Participants that Withdrew	5	5 M=4; F=1	0 M=0; F=0	4 M=3; F=1	0 M=0; F=0	1 M=1; F=0	0 M=0; F=0
Total Participants Involved in Wave One	31	17 M=8; F=9	14 M=7; F=7	9 M=5; F=4	7 M=3; F=4	8 M=4; F=4	7 M=4; F=3
Recruitment Wave Two	19	11 M=4; F=7	8 M=6; F=2	7 M=3; F=4	6 M=4; F=2	4 M=1; F=3	2 M=2
Participants that Withdrew	4	2 M=1; F=1	2 M=1; F=1	2 M=1; F=1	2 M=1; F=1	0 M=0; F=0	0 M=0; F=0
Total Participants Involved in Wave Two	15	9 M=3; F=6	6 M=5; F=1	5 M=2; F=3	4 M=3; F=1	4 M=1; F=3	2 M=2
Combined Recruitment Wave One and Two	55	33 M=16; F=17	22 M=13; F=9	20 M=11; F=9	13 M=7; F=6	13 M=6; F=7	9 M=6; F=3
Participants that Withdrew	9	7 M=5; F=2	2 M=1; F=1	6 M=4; F=2	2 M=1; F=1	1 M=1; F=0	0 M=0; F=0
Total Participants Involved in the Study	46	26 M=11; F=15	20 M=12; F=8	14 M=7; F=7	11 M=6; F=5	12 M-5; F=7	9 M=6; F=3

Table 2.2. The combined number of participants in waves one and two, showing the breakdown between the children with DS and TD children.

2.4 Materials

Once the participants were recruited and assigned to either of the experimental groups, packs containing instructions, two labelled boxes, eight salivettes, two labelled envelopes, and two pieces of tinfoil were delivered to the children's homes in the control group. Packs containing those items, as well as streamers, lollipop sticks, pipe cleaners, a kazoo, a tambourine, an egg shaker, rhythm family pictures and balloons, were also delivered to the homes of children in the group engaging with music (Table 2.3).

Control Group

Instructions	
Labelled Boxes	2
Salivettes	8
Labelled Envelopes	2
Tinfoil	2



Music Group

Instructions	
Labelled Boxes	2
Salivettes	8
Labelled Envelopes	2
Tinfoil	2
Streamers	4
Lollipop Sticks	4
Pipe Cleaners	6
Kazoo	1
Tambourine	1
Egg Shaker	1
Rhythm Family Portrait	1
Balloons	2

Table 2.3. The contents of the MINdDS bags for participants in the control and music groups.

The study took place over eight weeks with pre- and post-testing in both groups. Before the intervention commenced, saliva and hair were collected, while the Peabody Picture Vocabulary Test (PPVT-4) and cognitive tests were administered (Table 2.4).

Questionaires	Biological Samples	Age-Matching Test	Cognitive Tasks
Why Choose Music?	Saliva and Hair Samples	Peabody Vocabulary Picture Test	Temporal Order Judgement Task
PedsQL Quality of Life Inventory			Corsi Blocks Test

Table 2.4. The pre-test measures that each group required at the start of the study

2.4.1 Questionnaires

All questionnaires were administered through the Qualtrics survey platform to which parents were given a specific link. All participants completed two questionnaires at the start of the study. The first questionnaire entitled 'Why Choose Music?' (Paterson, 2008) (Appendix B) was only filled out at the pre-test stage to give the researcher an insight into the musical experience/background of both the parent/guardian and the child (Paterson, 2008). This questionnaire comprised five sections, with some requiring respondents to rank their responses on a Likert scale of 0–7. Open-ended questions were also included to allow the parent/guardian to elaborate on their answers. The questions in the first section focused on the demographic details of the parent/guardian. The questions in the second section explicitly concentrated on the musical experience of the parent/guardian. The third section was for those whose children were involved in Music4Children. The questions in the fourth section of the questionnaire were specific to those who were not engaged in music. Finally, a fifth section allowed the parent/guardian to add additional comments reflecting on their engagement, or lack thereof, in music and comment on its importance.

The Paediatric Quality of Life Inventory (PedsQL) relevant to the child's age group was also completed by the parents/guardians of both cohorts before and after the intervention. This gives an insight into the quality of life of a child (Varni, Seid, Rode, 1999). There were three versions of the questionnaire: one suitable for 2–4-year-olds, one for 5–7-year-olds, and finally, one for 8–12-year-olds (Varni et al., 2001) (Appendix B). Each form included 23 questions which directed parents to comment on their children's quality of life under the headings of physical functioning (8 items), emotional functioning (5 items), social functioning (5 items), and work/studies functioning (5 items). The questions were scored on a 5-point response scale ranging from 0 (never a problem) to 4 (almost always a problem) (Varni et al., 2001). The items were then inversely scored on a scale of 0-100 (0=100, 1=75, 2=50, 3=25, and 4=0). The total scores in each section were collated and divided by the number of items in each section. The mean value was then calculated from each score (Varni et al., 2001).

The group engaging with music completed two additional questionnaires. A Daily Diary that consisted of a series of multiple-choice questions was completed by the parents/guardians after their child engaged with the online video (Appendix B). The parents/guardians of the group engaging with music were also encouraged to complete the 'Qualitative Description of Your Child's Experience of the Music Intervention', for which approval was granted in March 2022 after the completion of wave one. Recruitment took place only after ethical approval was granted, following which the questionnaire was completed. This included rating their child's engagement in the intervention on a Likert scale from 1–10, while open-ended questions encouraging parents to give more details on their child's engagement with the music programme also featured. Details of the questionnaires are in Appendix B.

2.4.2 Saliva Collection and Storage

To assess if the music intervention had an impact on the concentrations of cortisol and oxytocin saliva was collected. The parent of each participant collected saliva samples in Sarstedt v-bottom salivette® tubes provided. Under guardian supervision, the child put a small sponge in their mouth for 2 minutes to absorb as much saliva as possible. This was then placed directly into the tube without being handled. Four samples were taken one week before the intervention phase: on waking, 15 minutes, 30 minutes, and 45 minutes after waking. The participants did not consume food or drinks to reduce contamination until all the samples had been collected (Bowling et al., 2022). These were then labelled to identify the times and dates at which samples were taken and included the subject ID of the participant. Salivette tubes were stored by the parent in the fridge at 4°C until collected and transported in a mobile fridge to the lab by the investigator. The samples were centrifuged at 4°C, 1500 x g, for 10 minutes and aliquoted into Eppendorph tubes. Three hundred ml was aliquoted into one tube for the analysis of oxytocin, while the remainder was aliquoted into another to analyse cortisol. All samples were frozen at -80°C until required for assay. The saliva collection was repeated at the end of the seven-week intervention for both the control and intervention groups. Those living outside of Cork were exempt from providing saliva samples as collecting the samples on the same day was impossible.

2.4.3 Hair Collection and Storage

Hair samples were also collected to measure the concentration of cortisol before and after the intervention in both cohorts. The parents/guardians collected 50–100 strands of hair at the posterior vertex as close to the scalp as possible, about 3 cm in length, as seen and as recommended by the Society of Hair Testing (SoHT) (Wenning, 2000; Carro et al., 2022). The portion of hair that was closest to the scalp was covered with tinfoil. The parents then placed the hair sample in the envelope provided and added their subject ID to the label. Hair collection was repeated one week after the intervention concluded. The investigator collected the hair samples from participants' homes in the county, whereas those living outside the county were asked to post the hair sample.

When processing the samples, 10mm of hair was cut from the end closest to the scalp using a ruler, and then the hair was cut into 1 mm minuscule pieces and weighed. The optimum weight required was 10 mg, then placed in small tubes and placed in a box labelled 'pre-intervention' or 'post-intervention' (Meyer et al., 2014). Hair samples were stored in a cool dark drawer until the analysis of cortisol concentration could be carried out. Cortisol was extracted by using methanol (Carro et al., 2022). The extract was then aliquoted, and the solvent evaporated. The extract was processed by automated chemiluminescent immunoassay (CLIA) Immulite 2000® with minor modifications (Gonzalez et al., 2019). Cortisol results were expressed in pg/mg of hair (Carro et al., 2022).

2.4.4 Peabody Picture Vocabulary Test

After being randomly allocated to each group, the children were MA-matched using the PPVT-4 (Dunn & Dunn, 2007). This is an assessment tool for mental age matching through evaluating receptive vocabulary widely used by clinicians and researchers. The lack of consistent assessment models before trials and interventions has often skewed results. As previously acknowledged, a review by Yang et al. supports the PPVT as a preferable way for MA-matching groups as it uses receptive vocabulary, which is more accessible to children and adults with DS (Yang, Conners et al., 2014).

The test was delivered online, where the investigator shared their screen. Each child was given a chance to answer a sample test first to ensure they knew what was expected; this was not marked. The child then pointed to their screen, and the parent identified which number the picture was. There were ten sets of pictures in total that ranged from 2.6 years to 11–12 years. The corresponding set appropriate to the child's age was selected for TD children. However, when testing the children with DS, it was advised to test from two years below their CA. To ascertain the bass level, the child in each cohort must have no more than one error in a particular set, and the ceiling set is achieved when the child has eight or more errors (Dunn & Dunn, 2007). Once a set is started, it must be completed. The raw score was attained by subtracting the number of errors from the ceiling item. The raw score was then converted to a standard score with a mean of 100 and a standard deviation of 15 (Dunn & Dunn, 2007). The corresponding percentile range, normal curve, and stanine score were attained to calculate the MA of each child with DS (Dunn & Dunn, 2007).

2.4.5 Cognitive Tests

At the start of the study, the participants in both cohorts were required to take two tasks, the Temporal Order Judgment (TOJ) task and the Corsi Blocks test administered on the Pavlovia platform. The platform permitted parents to access the tests through a laptop. These tests were devised to test short-term spatial memory and temporal perception.

To further increase the programme's accessibility, the children were introduced to Mert, a friendly mouse who had gone missing. The recorded narrative was activated as the child was introduced to a new test. The complete narrative may be found in Appendix D. It was based on a 3x2 mixed design Analysis of Variance test (ANOVA), with Repeated Measures Design with Modality (audio only, visual only and then audiovisual) with Stimulus-Onset Asynchrony (SOA) as factors.

The first section of the TOJ task consisted of an auditory stimulus with one beep appearing at the following time intervals: 0 ms (Stimulus Onset Asynchronies), 50 ms, 100 ms, 150 ms, 200 ms, 250 ms, 300 ms, 500 ms. To assist the children, the researcher asked them to "use their listening ears to find Mert, who was missing in the garden". Beeps were administered at either the left or right side, and the children

had to guess which side by pressing the cursor keys, the right arrow when they heard Mert on the right side, and the left arrow when Mert was heard on the left side. In the second phase, the children were presented with a visual stimulus. The children were told that they were "now using a flashlight to help find Mert", the light of which would appear above or below the crossroads. The left arrow was pressed when the light was above the road and the right when below. The third phase included the presentation of both auditory and visual stimuli, both flashes and beeps, at which the participant must decide which was presented first. Therefore, the children had to use their flashlight and listening ears to find Mert. There were 210 trials in the audio-visual block, which was evenly divided across all conditions, with 70 trials each. This was followed by the Corsi Blocks test, which is frequently used with people with DS, as their spatial memory is often less affected than other cognitive areas (Lanfranchi, Carretti et al., 2009). The participant was told that these were blocks of cheese and that they had to put them in the order they were presented. The Corsi Blocks test had ten trials in total. Both the control and intervention groups were required to repeat the cognitive test at the end of the intervention.

2.5 Materials and Procedure for Those Engaging in the Music Intervention

Children assigned to the music group engaged in a music intervention programme for seven weeks. In keeping with the literature, and as outlined previously, the word intervention is used in this context as the impact music has on the cognitive aspects of both cohorts is being evaluated. The programme was delivered via online videos and a weekly online music class, aided by a take-home pack. We did not account for music preferences. My experience of devising music programmes for Music4Children informed the content of the intervention. Therefore, the programme included modular elements embracing the main fundamental components of music, including pitch, rhythm, tempo, and texture, through singing, instrument playing/recognition, and various musical memory-based games.

The seven videos (one for each week) were 15–17 minutes long and watched Monday through Friday. They were supplemented with a 40-minute online class facilitated through MS Teams on Saturday. To increase flexibility, a choice of times at which children could engage with the online class was provided: 11 a.m. or 2 p.m.

The online class was designed to introduce the children to the material in the video scheduled for the following week. The parents were also present online to ensure the technology worked. The links to the videos can be found at the following YouTube locations (Table 2.5), while images from the intervention are shown in Figure 2.6.

Lesson	YouTube Location
Lesson One	https://youtube/za9u6O3_EEg
Lesson Two	https://youtube/kVKRaVI-BBY
Lesson Three	https://youtube/FidOAtutBZ8
Lesson Four	https://youtube/vvqcTAvkvb4
Lesson Five	https://youtube/MaRMnf82uXM
Lesson Six	https://youtube/QRk0BaSFwbU
Lesson Seven	https://youtube/DrfSlh4Z53I

Table 2.5. The YouTube location for the online classes. These videos remain unlisted on this platform, and links were only available to those engaging in the study.



Figure 2.6. Selected Images from the MINdDS Intervention.

The consistency of delivery was paramount, and an in-built lesson structure accommodated alterations to the content while maintaining a continuous routine, as seen in Figure 2.7



Figure 2.7. The lesson structure that was adhered to in each lesson in the MINdDS intervention.

Whichever mode of delivery was being used, the session would commence with the 'Hello' song. The initial song was followed by an introduction/discussion of the rhythm family members and familiarising them with their rhythmic value. Each member of the rhythm family was presented with a contrasting narrative. As the weeks progressed, the rhythm family were presented with varying degrees of difficulty on the face of an animated clock. These clocks were created in collaboration with students during active research before the intervention (Appendix E). The make-and-do section comprised making the rhythm family members out of pipe cleaners, providing the children with a visual they could retain in their sound bag and refer to from week to week, and, most importantly, bringing them to each lesson.

Contrasting musical concepts were introduced each week through fun and engaging songs with permission from MusicK8 and Plank Publishing. These included the dynamic markings of *piano* (*p*), pianissimo (*pp*), *forte* (*f*), *fortissimo* (*ff*) as well as *crescendo* (*cresc*.). In addition, the tempo markings *presto* and *largo* also featured, as well as ternary form. Each week, there was an instrument parade with six instruments presented in a particular order, which would disappear from the screen, and the children would have to remember them in the original sequence in which they

appeared. For example, these included a tambourine, egg shaker, sleigh bells, kazoo, agogo, and triangle. The parade was always followed by a sequential song, which changed every two weeks to give children ample time to learn it. A sequential song is a song that follows a particular pattern or sequence of events. This gives children a sense of what is coming next, allowing them to 'predict'. Each session would conclude with a Goodbye song. Every musical element/method was designed to activate specific brain regions, and a corresponding measure was used to assess its effectiveness, as shown in Table 2.6.

Brain Function	Musical Method	Brain Region	Measures
Learning and Memory	Sequential Songs Songs with instructions and movement Synchronisation Instrument Parade	Cerebellum Hippocampus Prefrontal Cortex	Temporal Order Judgement Task Corsi Blocks Test PedsQl Quality of Life Inventory
Sensory-Motor Skills	Rhythm Family Improvisation Instrument Playing Group playing Beat Synchronisation	Motor Cortex Cerebellum Parietal Lobe	Temporal Order Judgement Task Corsi Blocks Test
Emotion Regulation	Music-Evoked Emotions through song	Striatum Amygdala Hippocampus Hypothalamus	Cortisol PedsQl Quality of Life Inventory
Social Bonding	Group Singing Group Performance	Striatum Amygdala Hippocampus Hypothalamus	Oxytocin PedsQl Quality of Life Inventory
Executive Function	Musical Clocks Rhythm Family	Prefrontal Cortex	Temporal Order Judgement Task Corsi Blocks Test PedsQl Quality of Life Inventory

Table 2.6. An overview of brain function, the musical method, brain structure, and the measures used in the study. The Peabody test was not analysed statistically and was used as a descriptive as it is the normative score that is relevant to mental age.

2.6 Qualitative Data Analysis

All qualitative data was analysed using content analysis as advised by Hsieh and Shannon (2005). This approach is flexible for analysing data where literature on a particular subject area may be absent or scant (Cavanagh, 1997; Hsieh & Shannon, 2005). Conventional content analysis was used specifically in this study; as described by Hsieh and Shannon, it is appropriate when using open-ended questions and interview-style approaches to research. The process of this kind of analysis includes several steps. The initial phase of this research included reading all the data several times (Tesch, 1990; Hsieh & Shannon). Then, it was read word by word to acquire codes (Miller & Huberman, 1994; Hsieh & Shannon, 2005). Words that were related

to specific concepts were then highlighted. This step was followed by rereading the data, taking notes, and making first impressions. The codes were then sorted into categories. The codes were also clustered (Patton, 2002; Hsieh & Shannon, 2005). A coding tree was then used to help organise these codes into categories and themes. This approach was used for all two qualitative data collection points.

2.6.1 Statistical Analysis

The primary inferential analysis used was ANOVA. The responses from the TOJ and Corsi Blocks were collated in Excel and then imported into The Statistical Package for the Social Sciences (SPSS) to generate the data output. The quantitative data from the PedsQL inventories were calculated from the cumulative scores under the main headings of physical functioning (8 items), emotional functioning (5 items), social functioning (5 items), and work/studies functioning (5 items). The quantitative data from the diary entries was calculated using the last entry, day five of each week of the intervention, as this was the most consistently answered across both cohorts. Those who did not fill in the diary on this day were not excluded from the analysis (Li et al., 2015). Therefore, the last entry was used when a participant failed to complete a diary entry on day five of a given week (Li et al., 2015).

Results

While the intervention was delivered in two separate waves, the results are combined to present the overall impact of the programme.

3.1 Establishing a Recruitment Profile

The 'Why Choose Music?' questionnaire was required to be completed to obtain a recruitment profile of the participants. Out of 46 participants, 37 completed the questionnaire, 13 of whom were from the DS cohort and 24 from the TD group (Paterson, 2008). Therefore, the compliance rate was 80.4% across both waves. The questionnaire was predominantly filled out by female parents (78.6%), with only 8.1% of the responses recorded by male parents. The age range of the parents and guardians was limited to the older age categories, and therefore, no participant aged between 18 and 29 participated in the study. There were 29 (78.38%) aged between 30 and 49 years, while 8 participants were 50+ (21.62%). There was a larger proportion of older parents in the DS cohort (35.5%) compared to the TD group (21.7%). The educational background of the participants' parents/guardians showed that 24 (64.8%) of this cohort had achieved a postgraduate-level qualification. There were 8 (21.6%) that attained a degree, with 3 (8.1%) reaching QQL6, 1 (2.7%) that achieved a vocational certificate, and another a diploma (2.7%) (Table 3.1). The highest number of participants are working in education, with healthcare and industry being the second most common forms of employment.

Gender of Parent/Guardian	Frequency	Relative Frequency
Male	3	8.10%
Female	28	78.60%
Unanswered	6	16.20%
Age		
18–29	0	0
30–49	29	78.28%
50+	8	21.62%
Qualification		
Postgraduate	24	64.80%
Degree	8	21.60%
QQ Level 6	3	8.10%

Vocational Cert	1	2.70%
Diploma	1	2.70%
Total	37	
Profession		
Healthcare	7	18.90%
Education	12	32.49%
Researcher	4	10.10%
Accounting	1	2.70%
Industry	7	18.90%
Homemaker	5	13.50%
Student	1	2.70%
Total	37	

Table 3.1. Gender, Age, Qualification, and Profession breakdown of parents from the responses to the Why Choose Music? Questionnaire.

The total number of children involved in MINdDS after withdrawal was 46. There were 26 TD children (m=11 and f=15) and 20 children with DS engaged in the study (m=12 and f=8). Fourteen TD children (m=7 and f=7) and 11 children with DS (m=6 and f=5) received the music intervention. Twenty-one children acted as the control group (m=11 and f=10); of these, there were 12 TD children (m=5 and f=7) and 9 children with DS (m=6 and f =3). When considering the gender divide between the children involved in the study, it was found that 20 were male and 17 were female. While almost an equal division between genders was found in the TD cohort, there were over twice as many male participants as females in the DS group. The gender divisions between the two groups may be found in Table 3.2.

Waves One and Two	Total Number of Participants	TD n	DS n	Music Intervention TD n	Music Intervention DS n	Control (No Music) TD n	Control (No Music) DS n
Total Participants Involved in the Study	46	26 M=11; F=15	20 M=12; F=8	14 M=7; F=7	11 M=6; F=5	12 M-5; F=7	9 M=6; F=3

Table 3.2. The number of children involved in the study, including the gender breakdown between TD and DS cohorts across the experimental and control groups.

The PPVT-4 was used to test receptive vocabulary and to establish an exact MA-match for the children with DS (Dunn & Dunn, 2007). The PPVT-4 has been used extensively in MA-matching and is effective, mainly when working with children with DS, as their receptive language is less impaired (Yang et al., 2014). The recruited TD children were aged between 4–8 years. Everyone in the TD cohorts scored their age

equivalent or above, while the DS cohort scored considerably lower than their age range, with some participants being untestable.

Figures 3.1 and 3.2 illustrate the MA-match (blue bars) compared to their CA-match (orange bars) in the two DS cohorts (those engaging or not engaging with music). Both graphs display the age range of individuals with DS and the apparent differences between their CA and MA.

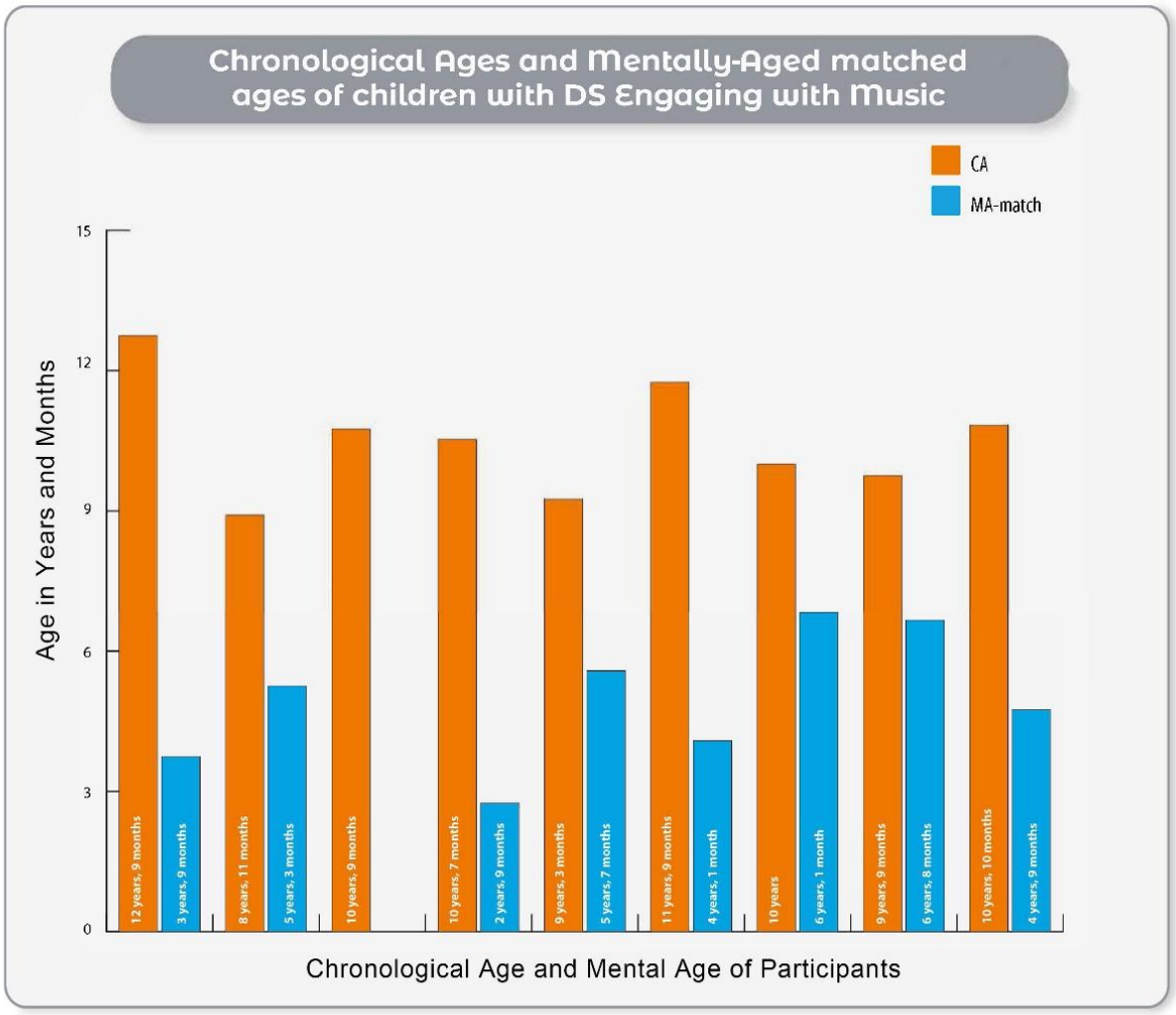


Figure 3.1. A comparison between the chronological ages of the children with DS ***engaging*** with music at the time of the study to their MA-matched ages derived from the PPVT-4 results.

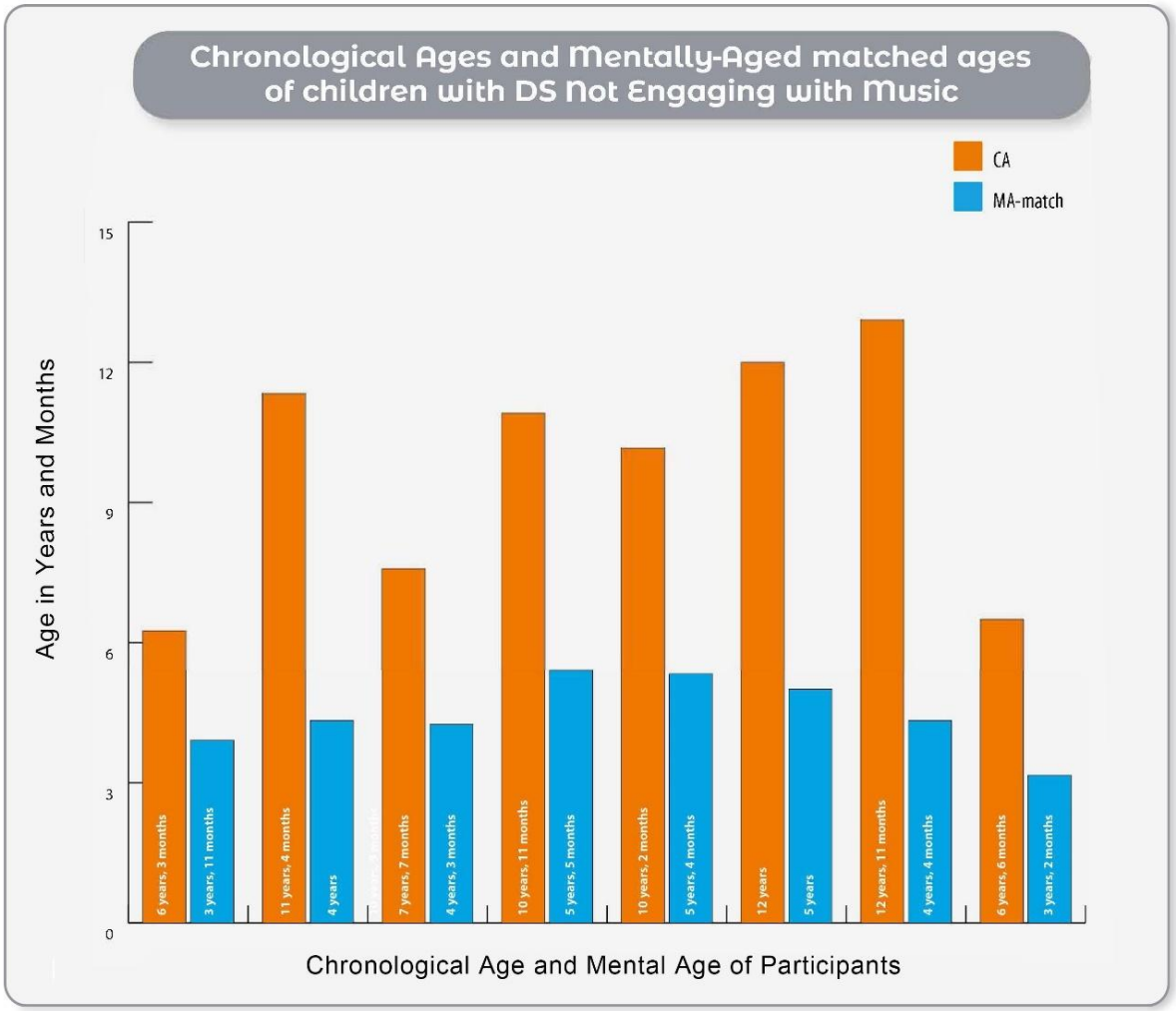


Figure 3.2. A comparison between the chronological ages of the children with DS ***not engaging*** with music at the time of the study and their MA-matched ages derived from the PPVT-4 results.

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A two-tailed T-test was carried out to see if there was a significant difference between the CA of the children with DS in comparison to their MA (p <.0001). The minimum CA of this cohort was 6.25 years with a MA of 2.75 years, while the maximum age was 12.91 years with a MA of 6.6 years. The median CA was 10.37 years, while the MA equivalent was 4.54 years, as seen in Table 3.3 and Figure 3.3.

Variables	Number of Values	Mean	Standard Deviation	t-score	DF	Sig. level (p)	Remarks
CA	16	10.09	2.009	9.898	15	<.0001	Sig.
MA	16	4.649	1.059				

Table 3.3. This table shows that the mean CA was significantly higher than the mean MA in the participants with DS, both engaging with music and not engaging with music (t=9.898; df=15; p <.0001).

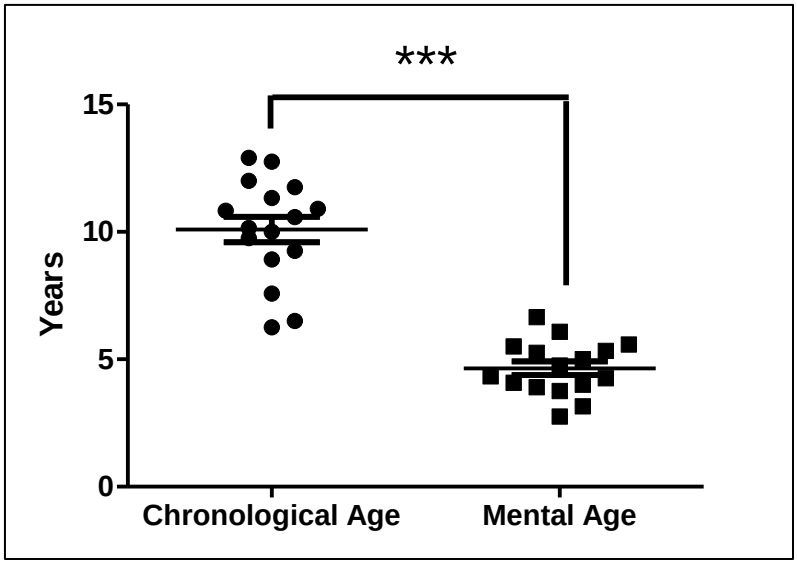


Figure 3.3. T-test showing a significant difference between CA and MA of the DS cohort (p<0.001; n=16).

3.2 Previous Musical Experience and the Perceived Importance of Music

An additional section of the 'Why Choose Music' questionnaire aimed to establish what previous experience, if any, the participants and their children had with music. The questionnaire was also used to ascertain what value they put on music and the impact it can have. Reflecting on their engagement with music, 62.16% of the parents/guardians always listened to music. However, only one guardian stated, 'it was very true of me' that they attended concerts, with 24.32% believing this

statement was ‘not true of me’. It was found that 78.3% do not play any musical instrument, with 2.7% saying the statement was ‘very true’. This coincided with whether they considered themselves to be a musician, to which 86.49% stated that this was ‘not true’ of them, while 5.41% stated this was ‘very true’. Of the 37 participants, 2 (5.4%) had been paid professionally to teach or perform music. The breakdown between the cohorts shows that they are evenly matched in their responses, as seen in Table 3.4.

Question	DS Mean	DS Count	TD Mean	TD Count	Total Mean	Total Count
I listen to music regularly (0 being 'Not at all True of Me' and 7 being 'Very True of Me)	5.50	14	6.17	23	5.92	37
I regularly attend concerts (0 being 'Not at all True of Me' and 7 being 'Very True of Me)	2.79	14	2.91	23	2.86	37
I play one or more musical instruments. (0 being 'Not at all true of Me' and 7 being 'Very True of me)	1.51	14	1.96	23	1.81	37
I consider myself a musician (0 being 'Not at all true of Me' and 7 being 'Very True of Me)	1.07	14	1.70	23	1.46	37
I am being paid to teach or perform music. (0 being 'Not at all true of Me' and 7 being 'Very True of me)	1.21	14	1.61	23	1.46	37

Table 3.4. The measures of central tendency of the parents who engaged with Why Choose Music? collectively, in both TD and DS cohorts.

Those whose children had engaged with a music programme, in this case, Music4Children, were then encouraged to move to the next section of the questionnaire, and those who had never engaged with music were prompted to go to the last section. Thirty-five of the 37 participants proceeded to either section; of

these, 8 (4 TD children and 4 children with DS) had previously enrolled their child in the Music4Children programme, therefore 22.8% of the cohort. There were 62.5% of this music group who wanted their children to experience a wide range of activities, and there was no motivation for improving their child's musical skills. When asked if they participated in a music programme because their child's friends were involved, 100% of the cohort stated this was 'not true at all of me'. It was found that improving the discipline of a child was not a motivational factor for their involvement. Eighty-seven per cent of parents stated that their children did not ask to be involved in music. Only one parent believed that music would improve their child's social skills, with 42.5% believing music was very important for all children.

Those who had never been involved in music proceeded to section four of the questionnaire, of which there were 27 (77.1%) of the 35 participants. When asked if their child had refused to participate in a music programme, 87.5% said this statement was 'not at all true of me'. Similarly, 26 out of 27 participants strongly believed that music was a valuable activity to be involved in, with 92.59% strongly disagreeing with the statement that their child would not enjoy music or that they do not have musical talent. The participants also responded positively to their child showing interest in music. Regarding the lack of knowledge on where to access the programmes, 19.2% stated this was 'very true of me'. The expense of engaging with music was not a deterrent, with 76.9% of the group stating this was 'not at all true of me'. Similarly, the lack of opportunity was not a factor as to why their child has not availed of music lessons, with only 7.69% marking number 6 on the Likert scale. The question of a child's age and that they were too young was also less of a deterrent for parents, with only 25.93% stating that this was 'very true of me'.

The remaining questions in the survey allowed parents to explicitly state why they wanted their child to be involved in the music intervention or why they had not engaged with music up to this point. The questions also allowed them to express the value they felt music had, or could potentially have, on their children's lives. Table 3.5 shows the open-ended questions.

- **Section Three:** (Engaged with Music4Children). Please state any other reasons or comments as to why your child is engaged with music**.**

- **Section Four** (Not Engaged with Music): Please state any other reasons or comments why your child has not engaged with music.
  Section Five: Do you feel that the reasons for involving your other child/children in music programmes were any different from those of the child discussed?
- How important do you think music is in a child's early experiences?
- Please share any other thoughts or comments.

Table 3.5. The Open-ended Questions in the Why Choose Music? Questionnaire.

The answers were coded using the content analysis detailed in the methodology section, a sample of which may be seen in Appendix C. Figure 3.4 provides a breakdown of the themes and categories that emerged from this analysis.

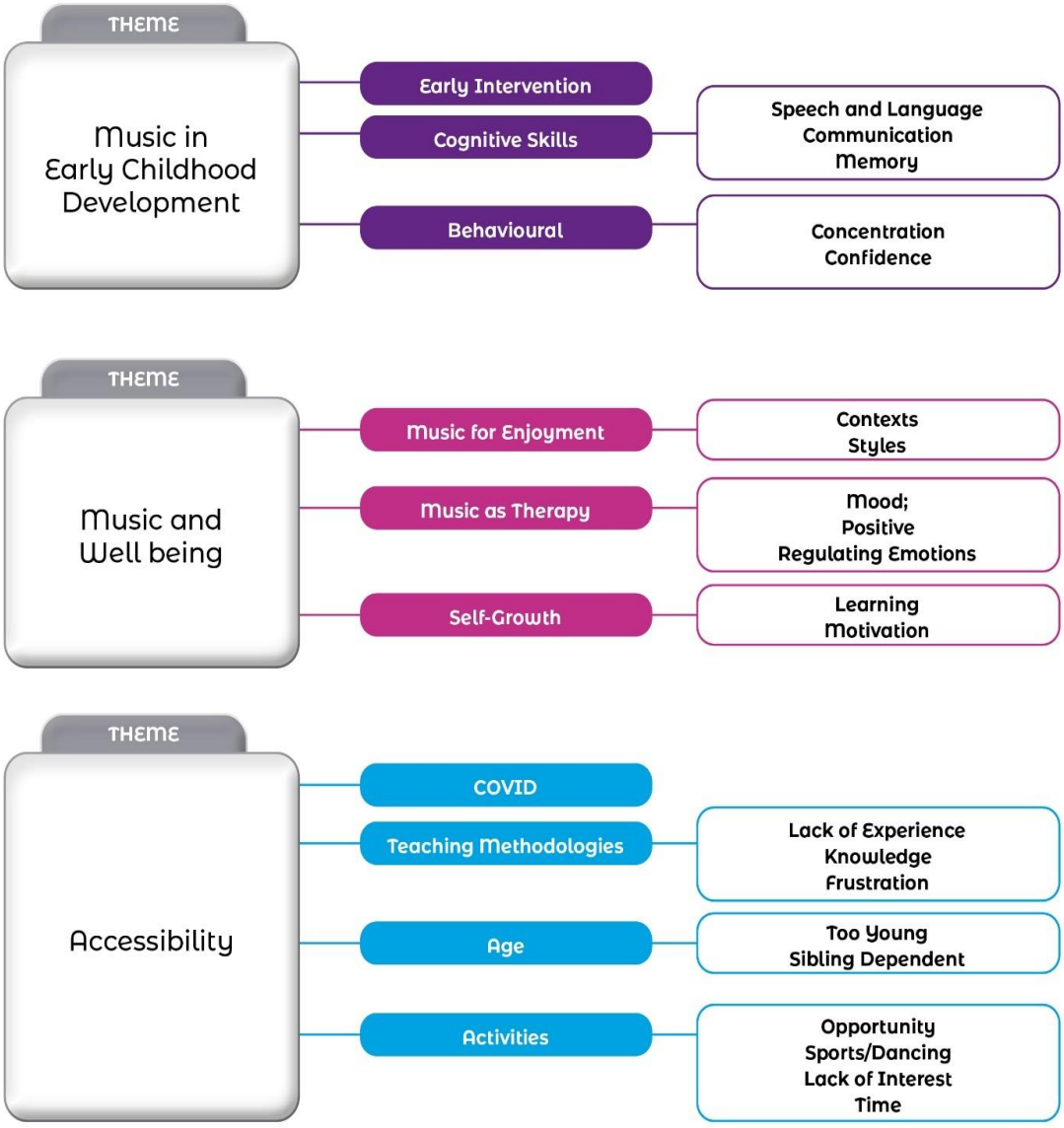


Figure 3.4. The themes, categories, and subcategories that emerged from the qualitative data obtained from the questionnaire, Why Choose Music?

3.2.1 Music in Early Childhood Development

The role of **Music in Early Childhood Development** was one of the main themes to emerge from the questionnaire, with one participant stating that music is "becoming increasingly important in early development." In line with this, the significance of music in an **early intervention** context was also highlighted, as it would help "to encourage interaction and communication." This was additionally acknowledged by a parent who believed that music should be introduced at an "early stage."

Under this theme of **early childhood development**, one of the categories to develop was the impact music has on **cognitive skills**, specifically **speech and language**. One parent felt that "music is extremely important for **speech**," which was echoed by another parent who "feel[s] music is very important as it contributes to **speech**." Similarly, an additional parent stated, "despite poor speech, [their] child is quite musical and sings in tune." The link between music and **language** was compounded in one participant's response: their "child responded to music from a very early age and could learn **language** in conjunction with music."

A related subcategory identified was **communication**, and music was viewed as "very important as another way of **communicating**." This was echoed by a parent who similarly viewed music as "a language in itself [that] aids **communication**." Likewise, an additional parent recognised that "music was **communication** amongst other things," while another acknowledged that, in fact, music "was another way of **communicating**."

The potential impact of music on **memory** also received emphasis in the questionnaire responses. While stressing the importance of music, one parent stated that it was essential for "**memory** and recall." One household had seen the benefits of "music and singing for both of [their] children for enjoyment, language learning, speech, diction, **memory** and winding down for bed."

The potential impact of music on behaviour was also recognised by parents, specifically in relation to **concentration** and **confidence**. There was a resounding response regarding the impact music has on **concentration**. One parent stated, "music is very important for speech, mood, **concentration** and memory." Several participants echoed this by indicating that music "helps concentration." **Confidence**

was also considered a positive response to music engagement, with one participant saying, “it promotes self-expression and **confidence**.” An additional participant stated that it “gives **confidence**, helps literacy, speech, social opportunities. fun.” Another parent is hoping “that music will encourage **confidence**.”

3.2.2 Music and Well-Being

Music and well-being were identified as intrinsically linked to music engagement. One participant felt that it “benefits personal **well-being** and general happiness.” When asked why they chose music, one parent stated it was for the **enjoyment** element that music provides. This **enjoyment** was linked to responses found in several subcategories as it “would be [the] main reason to encourage [their children’s] interests.”

The **contexts** of this **enjoyment** differed across the cohorts, as “there are a number of ways in how music can be integrated into a child's life.” This point was echoed by another participant stating their child enjoyed it at ‘school’, while others enjoyed the instrumental classes they attended, such as ‘guitar’. Others enjoyed music at ‘singing’ and ‘dancing classes.’ Another individual “sees how much happier [their children] are watching/listening to music.” Others felt that while they do not play instruments, they “have music in the house all of the time, and the children love music.” The need for music to be introduced in different settings and **contexts** was emphasised, but the need for variance in musical **styles** was also suggested. One parent stated that “all children should have the opportunity to be exposed to as wide a range of music as possible [and that they] would like [their] kids to enjoy music without feeling limited to any particular genres.”

Another emergent category from the responses was **self-growth**, with one parent stating that they felt music provided their child with ‘positive motivation’. This was also reflected in an additional response where the parent believed music was “for involvement and **self-growth**.” One parent felt music “elicits positive feelings; it stimulates.” Similarly, this was reiterated when a participant acknowledged that music “is imperative to [a child’s] development in a number of ways.” Another person mentioned how “music stimulates the brain and makes learning fun.”

One prevalent category that arose under **music and well-being** was **music as therapy**. One participant said that their "eldest found playing the piano **therapeutic**." Other parents believed that "music is extremely important for **mood**", while another 'cheered up' when listening to music. This was echoed by a parent who found that music can "help self-regulate **mood**." In fact, one parent "used music as a way to decompress with [their] daughter when she was very young. Before she learned how to regulate her big feelings." This was also reflected in a comment by a parent who found that "they would turn on some music and dance with [their child] if [they] felt she was angry or frustrated and could not find the words." Another parent stated, "music is a great way to express feelings...it resonates emotionally, for some, expression through movement to music works." The acknowledged role of music in relaxation was also found, with one parent finding that "music has featured fully in my child's life and has helped him stay relaxed and calm."

3.2.3 Accessibility

The **accessibility** of music was one of the prevailing themes identified as hindering people's ability to engage in musical activities. One of the main preventatives was the lockdowns associated with the **COVID-19 pandemic,** with many acknowledging it impacted their ability to find a musical outlet and "has added to uncertainty around signing up for activities generally." There were also "fewer options" due to **COVID-19**. In addition to the pandemic, some participants felt that there were few opportunities, with one participant citing that the "times and location of music classes [were] not available and none specifically for children with DS; hence [there was] little opportunity to learn." Another stated that while they had attended Music4Children "for a few sessions…the schedule did not really suit our family." Another noticed that they found increased availability for their TD child "to access music programmes much easier locally."

One resounding theme from the questionnaire analysis was the lack of appropriate **teaching methodologies** for children with DS. Some parents felt there was a massive **lack of experience** among teachers when teaching music to these children. One participant stated they were "concerned that the tutor would **not be experienced** in teaching a child with an ID." Another parent voiced their concern and

frustration when tutors do not possess the teaching resources or appropriate methodologies to work with a child with DS. They stated, "different teaching methodologies are important for children with an ID, and many tutors are unaware of this leading to **frustration** on all sides!" This was echoed by another parent who stated, "repetition is extremely important for children with an ID; often, tutors want to move on too quickly, which leads to **frustration**."

The accessibility of music programmes also depended on the child's **age** and whether they engaged in music. Some felt that children would benefit more if they were older playing an instrument, as one felt their child was "too young and would take up an instrument quicker when she is older." This was similarly uttered by another parent who stated that their "child has not tried an instrument as [they] feel she would pick it up quicker when she is a little bit older." Whether someone engaged with music depended on **siblings** as some were "close in age and generally attend activities together." Another parent highlighted that the "other children have no interest in music lessons"; therefore, they had not pursued lessons for their other child. From the opposite perspective, some would engage with music because their **sibling** was learning instruments. One participant highlighted that their "eldest plays piano and did play ukulele, has a DJ kit and loves to listen to music".

The **number of family activities** also impacted on people's ability to engage with a music programme. One participant said their daughter "is involved in many **activities**, including a dance class…" Similarly, another parent stated that their child "attends other **activities** and [is] conscious of not overfilling their weekends." Another also indicated they were "doing a lot of **activities** already." The main activities highlighted were sports, with one reporting that "the other children are very sporty and that [it] is all-consuming for them (so they do not make music)." Other parents felt that their children just had a lack of interest with their "other children hav[ing] no interest in music lessons although [they] have been offered various instruments". Others were conscious of the time required to dedicate to another **activity**, as it would require a lot of "extra time…for our kids to get going with a new activity."

3.3. The Experiential Impact of the Intervention

After the initial consent forms and questionnaires were completed, the children were randomly assigned to one of the four groups: the TD group not engaging with music, the DS group not engaging with music, the TD group engaging with music and the DS group engaging with music. The participants engaging with music were required to look at an online recorded class Monday to Friday and then attend an online live class on a Saturday. As seen in Figure 3.5, everyone attended the live class in lesson one in wave one, with a decrease in attendance from week to week. The lowest attendance was 67% (n=9) at lesson six, with 11 being the average number of attendees.

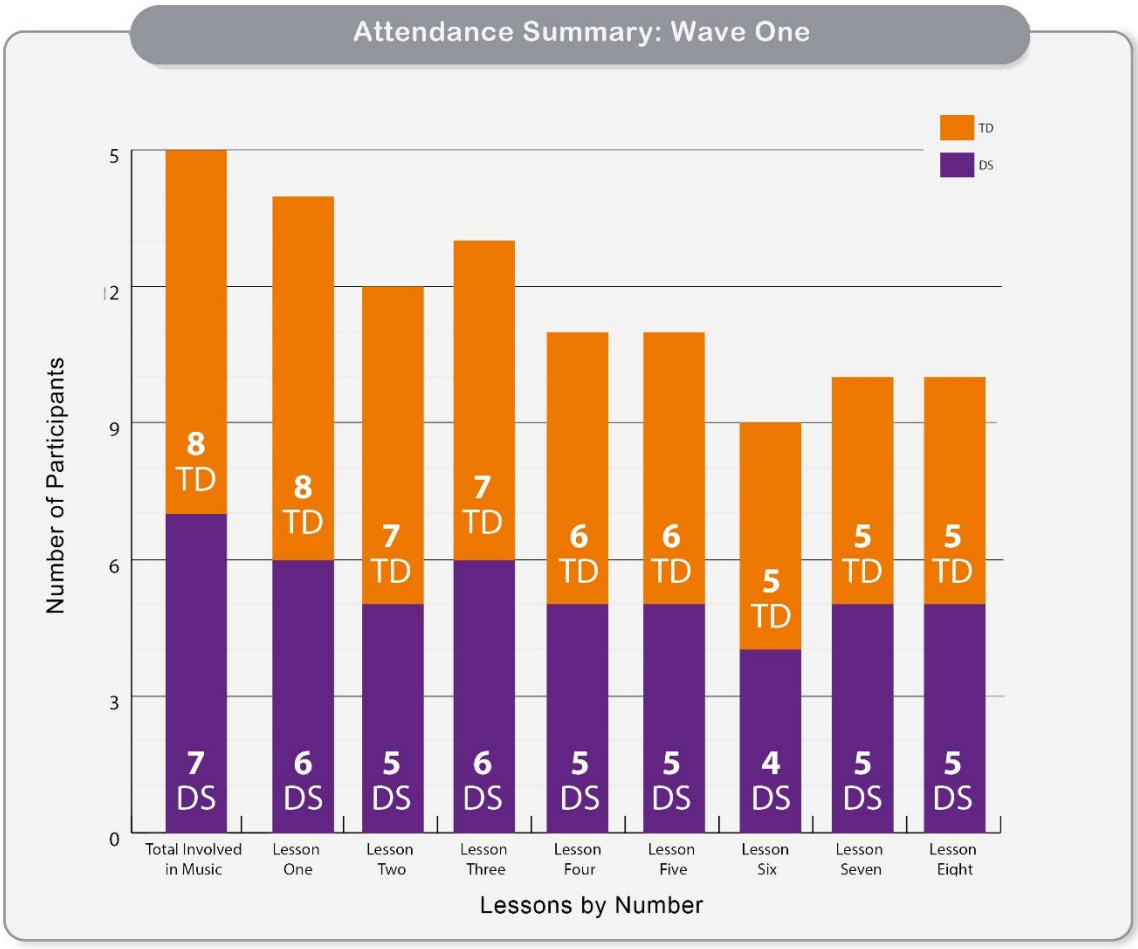


Figure 3.5. Attendance at the weekly lessons showing the DS and TD divide in wave one.

In wave two, nine were involved in the intervention group: 4 children with DS and 5 TD participants. There was no total attendance during wave two; the average

attendance was 7. The highest attendance at any lesson was 8(89%), and the lowest was 5 (56%), as seen in Figure 3.6.

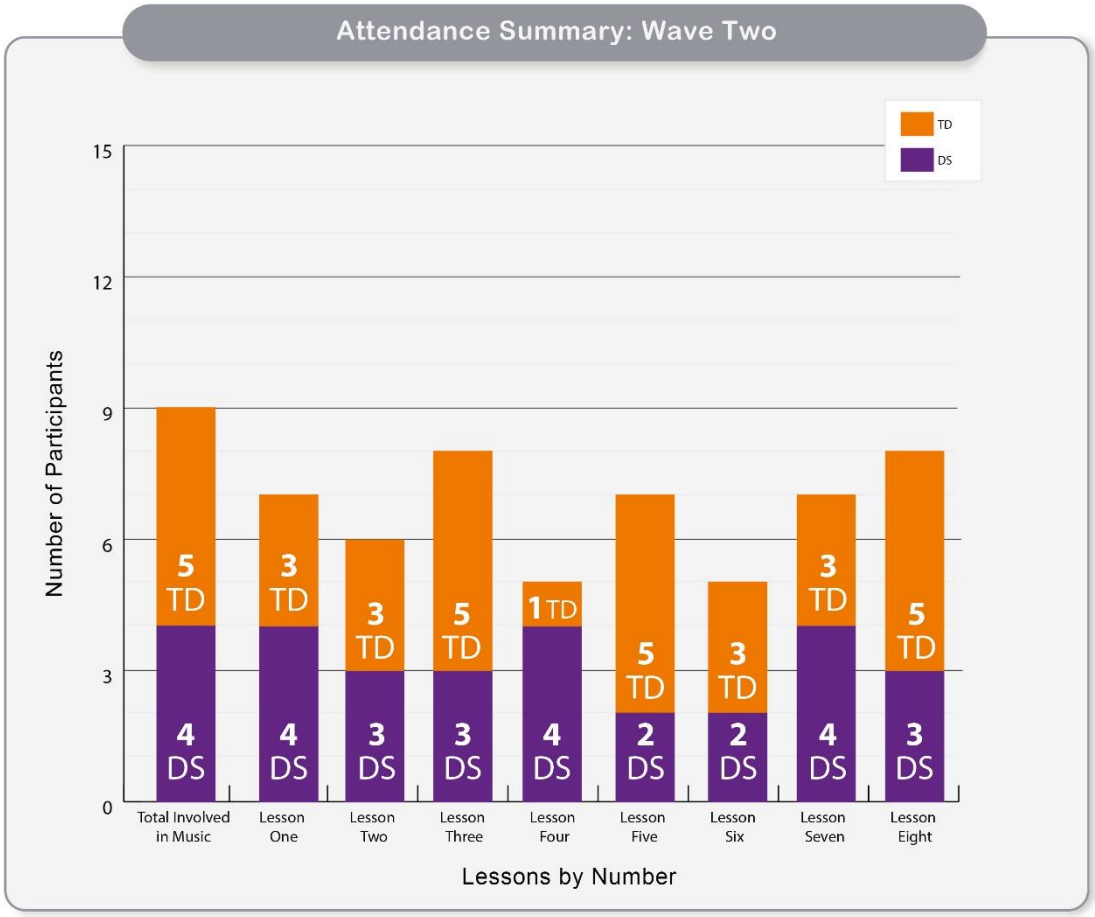


Figure 3.6. Attendance at the weekly lessons showing the DS and TD divide in wave two.

Figure 3.7 shows the total compliance between both waves. Lesson six was least attended by the children with DS, whereas the TD group began to disengage with the online class from lesson four.

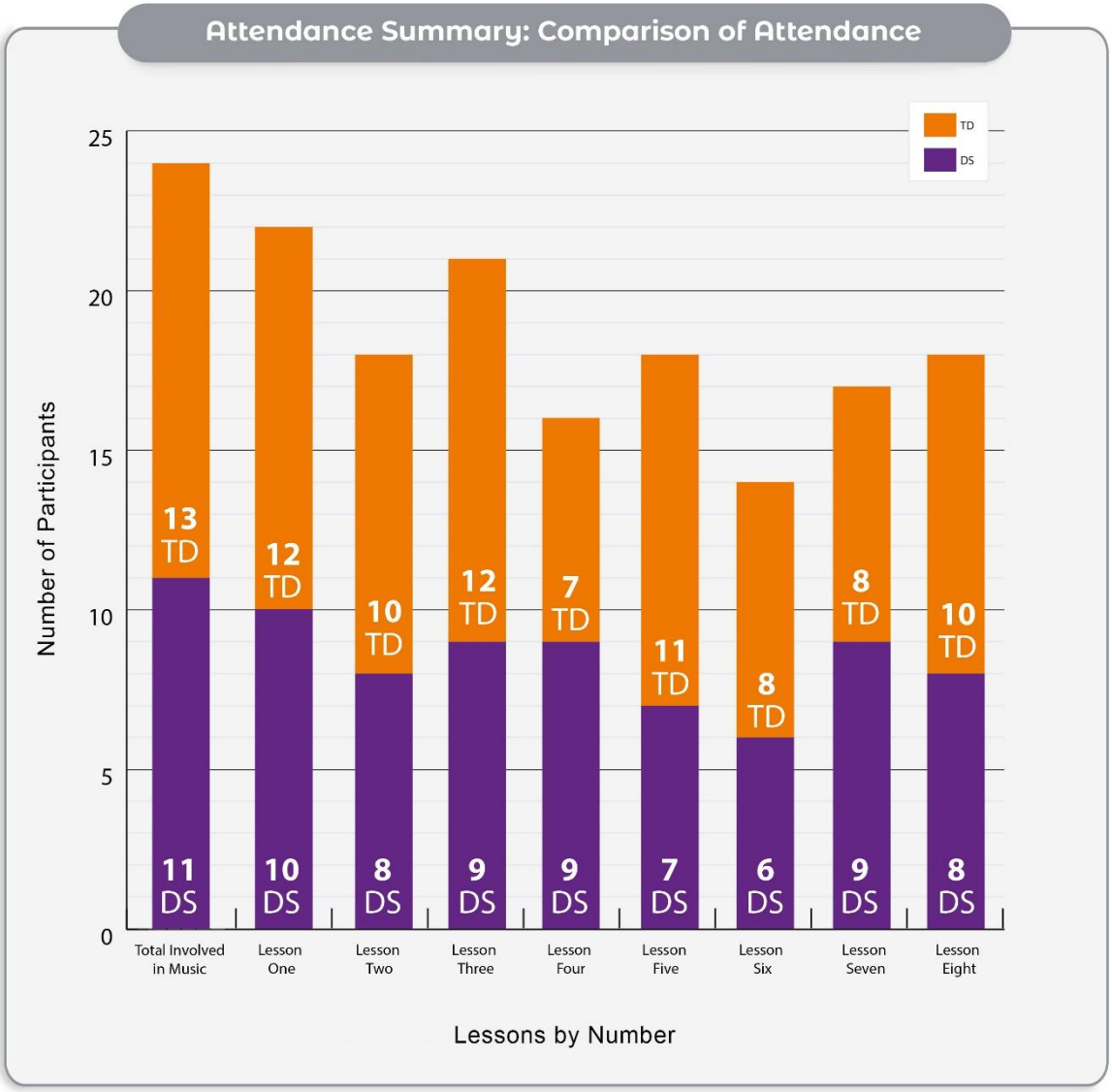


Figure 3.7. Attendance at the weekly lessons showing the DS and TD divide in waves one and two.

3.3.1 Daily Dairies

Each parent/guardian involved in the music intervention group was required to fill out a 'daily diary' (5 out of 7 days) to help note their child's engagement and progress with the music videos. Within this diary, they were asked to state how much time they engaged with the video that day and to rank their child's interaction with the sequential songs, instrument recognition, instrument parade, and music note recognition. There were 640 diaries recorded over both waves out of a potential 910. Therefore, compliance with the diary entries was 70%. The division of compliance between waves one and two may be seen in Table 3.6.

Wave	Frequency	Total Compliance Required	Relative Frequency
Wave One	362	560	64.6
Wave Two	268	350	76.5
Both Waves	640	910	70%

Table 3.6. Level of compliance with the Daily Diaries amongst both cohorts in waves one and two.

The participants were asked to specify how long their child engaged with the video, from 5 to 15 minutes, and equally encouraged to state if they did not look at it at all that particular day. Of the 640 responses, 75.63% engaged with the video for 15 minutes, 11.25% engaged for 10 minutes, 5.16% engaged for 5 minutes, and 5.16% did not engage with the video. The time spent engaging with music videos across cohorts may be seen in Figures 3.8 and 3.9.

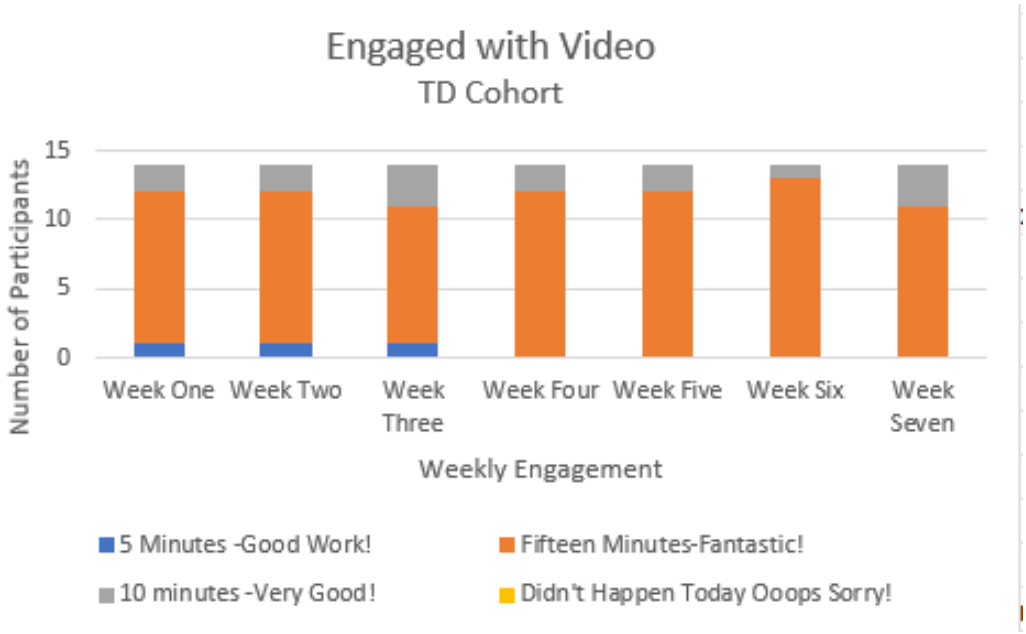


Figure 3.8. The time spent by the TD cohort engaging with the videos over the period of the intervention. The graph shows that most TD children engaged with the video for 15 minutes.

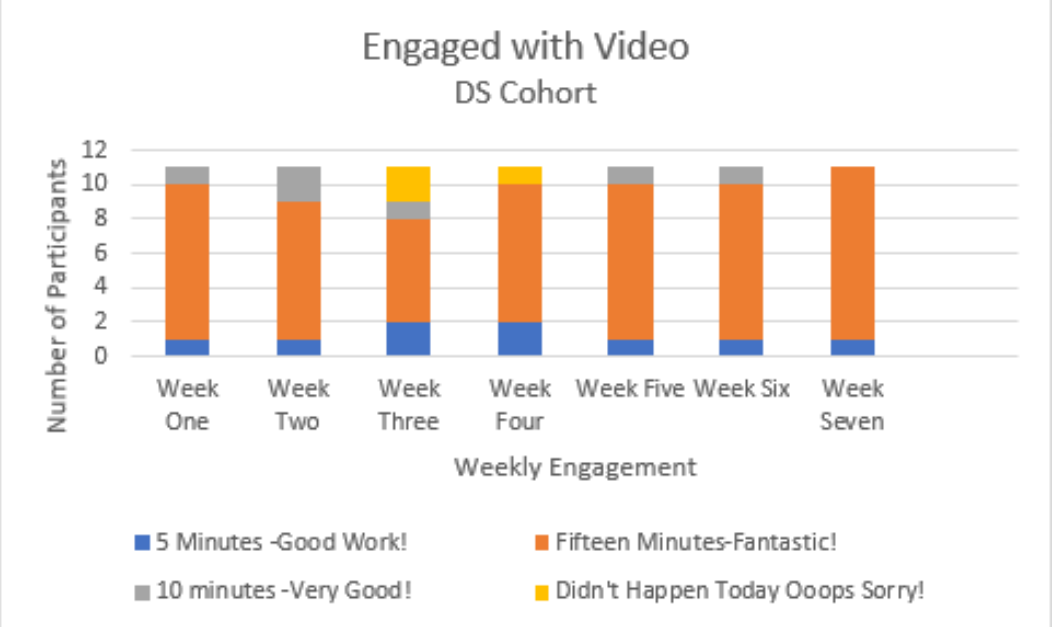


Figure 3.9. The time spent by the DS cohort engaging with the video throughout the intervention. The graph shows that most DS children engaged with the video for 15 minutes. However, in weeks three and four, some children who did not engage with the video.

Parents/guardians were asked how their child engaged with the sequential song. The importance of sequencing in early childhood has been acknowledged in the literature (Gouldthorp et al., 2017). A sequential song is, therefore, an extension of this idea that the song follows a set of consecutive actions and words that can be easily learned and predicted. The preferred interaction was through song and actions, with 76% of parents noting their child did both. However, 19% engaged with the actions only, while 13% observed. The number of TD children and children with DS engaged in both singing and actions, singing only, actions only, observing or made no attempt may be found in Figures 3.10 and 3.11.

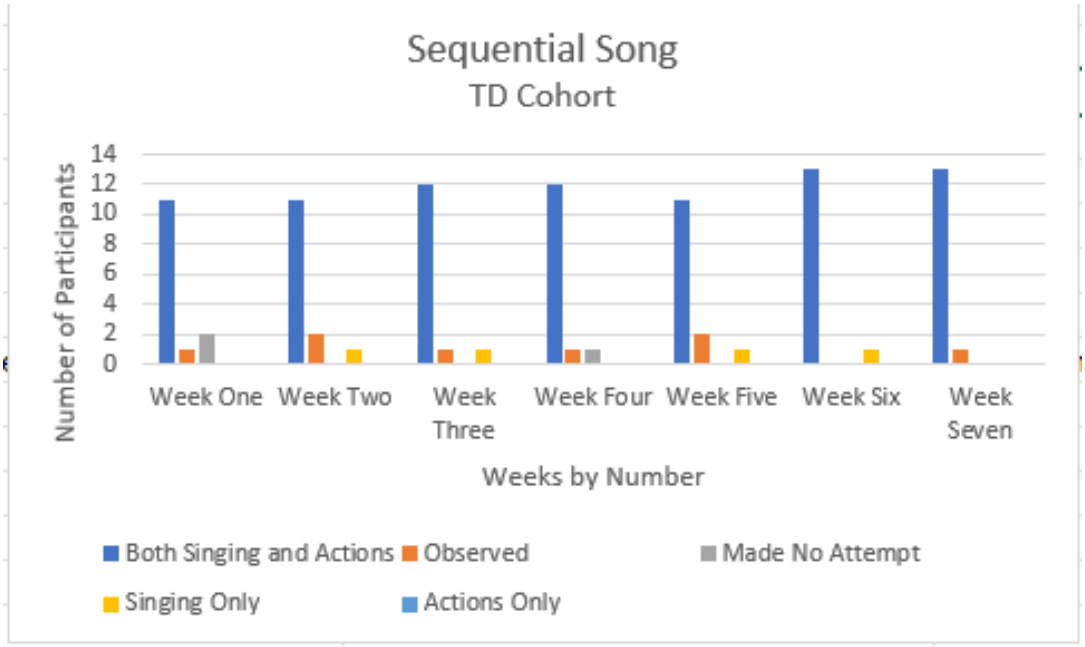


Figure 3.10. The graph presents the TD cohort's engagement with the video from week one to week seven. It identifies that the preferred engagement was both singing and actions.

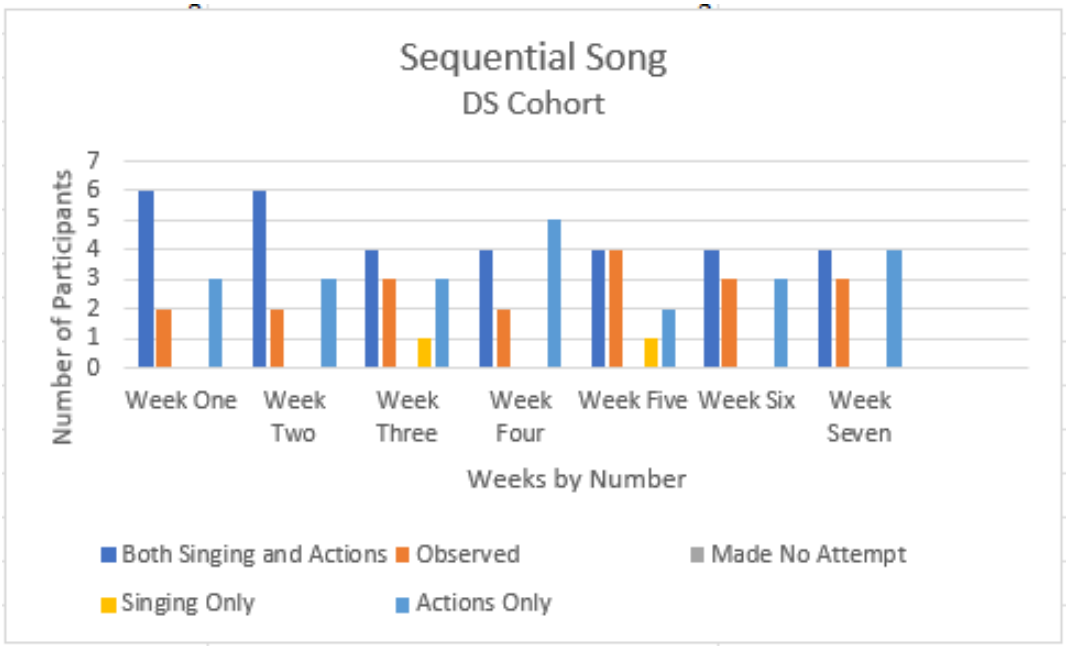


Figure 3.11. The graph presents the DS cohort's engagement with the video from week one to week seven. It identifies that the preferred engagement with the song was both singing and actions. However, by week seven, there was a comparable engagement with actions only.

Parents/guardians were asked about their child's ability to recognise the instruments. Of these, 64% recognised all, 21% attempted with some success, 8% made no attempt, and 7% attempted but struggled. Figures 3.12 and 3.13 show the number of TD children and children with DS who attempted but struggled, attempted with some success, recognised all, or made no attempt.

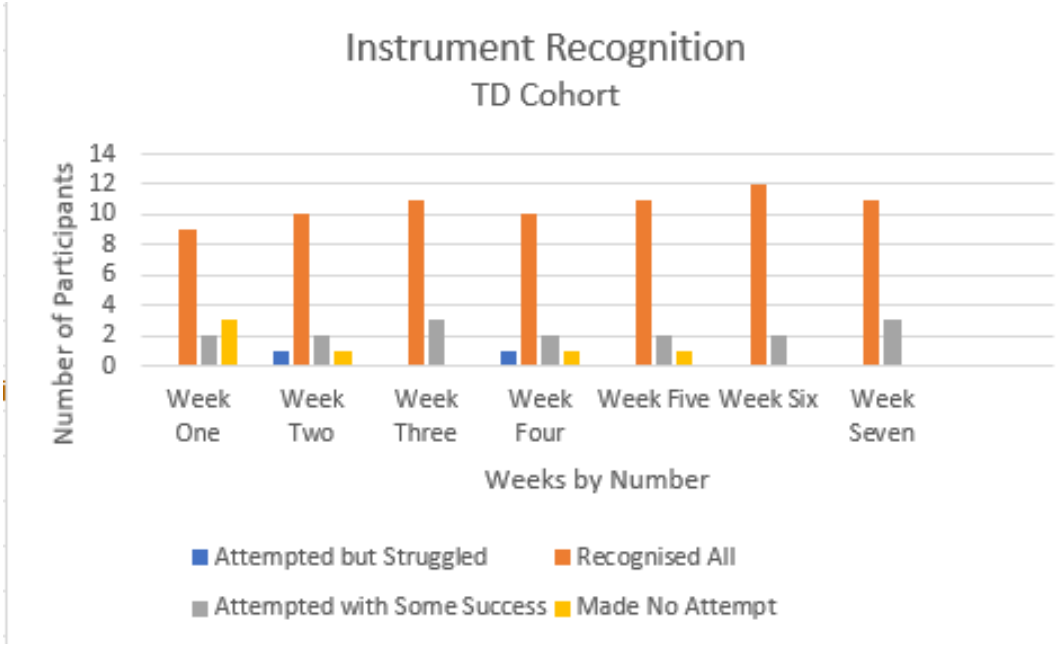


Figure 3.12. The graph shows the number of TD children who engaged with the instruments and the level to which they were successful in their recognition from weeks one to seven. Week six was the week the TD cohort was most successful.

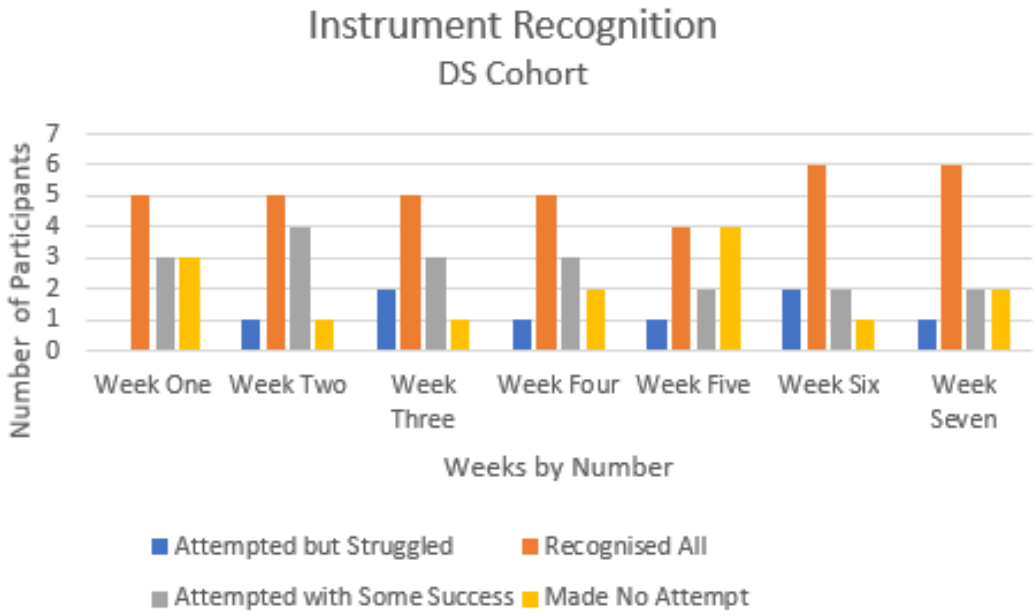


Figure 3.13. The graph shows the number of children with DS who engaged with the instruments and the level to which they were successful in their recognition from weeks one to seven. The DS cohort was most successful in weeks six and seven.

The children were presented with several instruments in a parade, of which they had to recognise the recurring order and identify the one that was missing. There were 62% that recognised all, 23% attempted with some success, 13% made no attempt, and 2% attempted and struggled. The exact number that recognised all, attempted

with some success, attempted but struggled, and made no attempt is found in Figures 3.14 and 3.15.

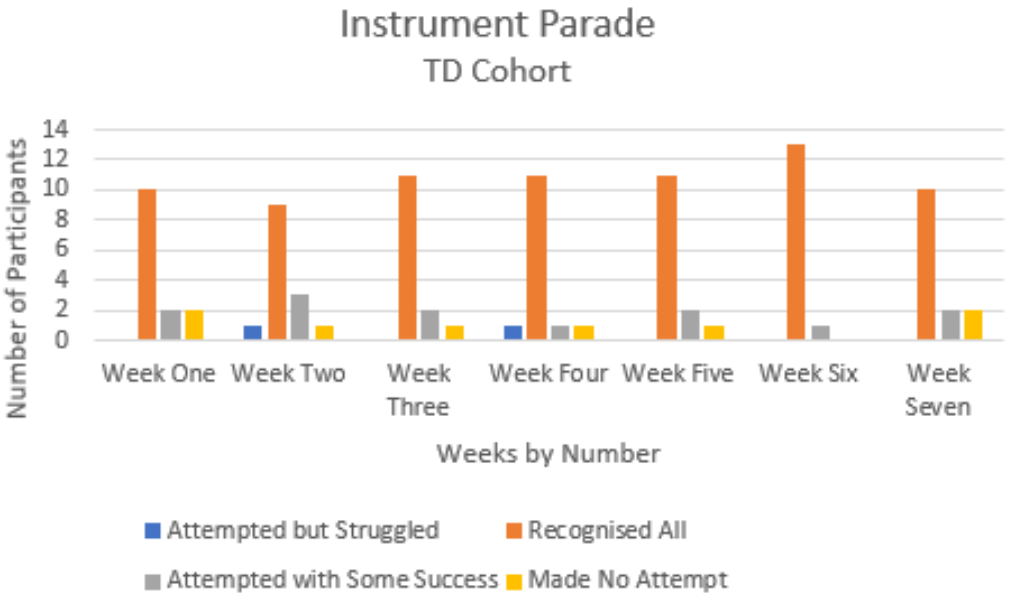


Figure 3.14. The number of TD children who engaged in the instrument parade and the level to which they were successful in their recognition from weeks one to seven. Week six was the week where the TD cohort was most successful.

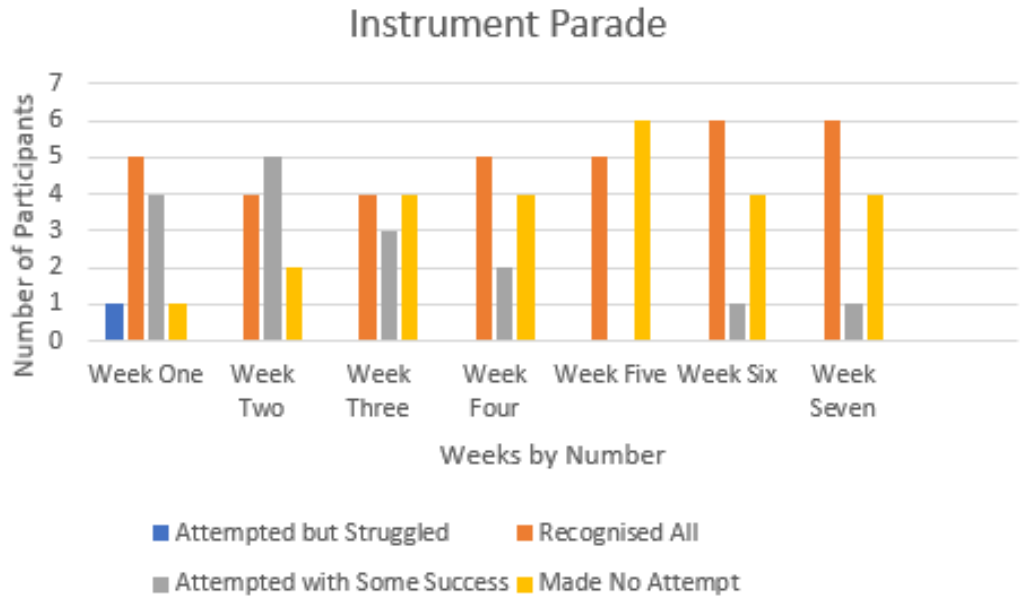


Figure 3.15. The number of children with DS who engaged in the instrument parade and the level to which they were successful in their recognition from weeks one to seven. Weeks Six and seven were the most consistent weeks for the DS cohort.

Becoming familiar with the melodic and rhythmic notes was integral to the intervention. There were 43% of children who recognised all the notes, while 37%

attempted with some success, 15% made no attempt, and 5% attempted but struggled. The exact number that recognised all, attempted with some success, attempted but struggled, and made no attempt may be found in Figures 3.16 and 3.17.

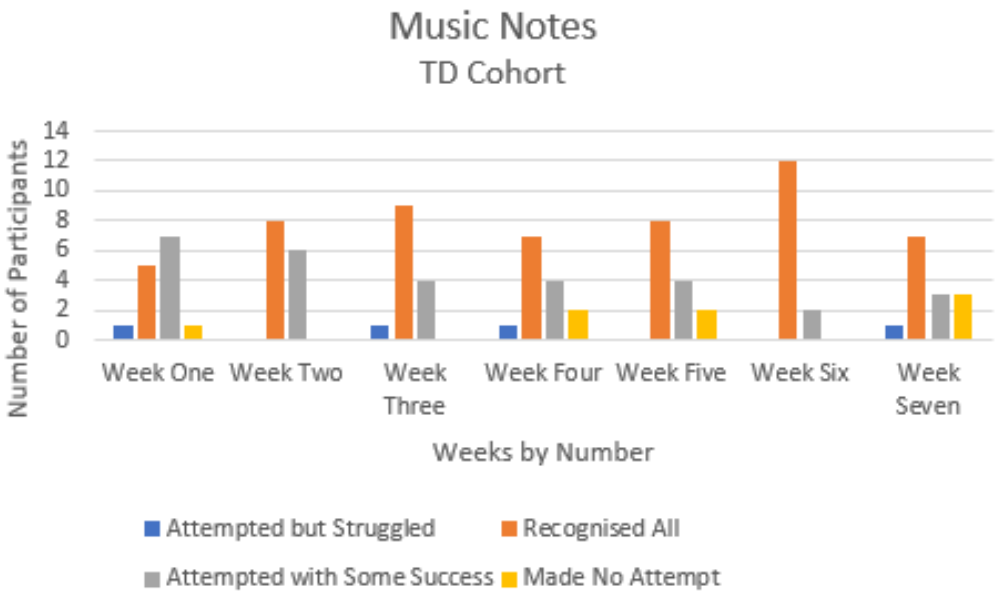


Figure 3.16. The number of TD children who engaged with the music notes and the level to which they were successful in their recognition from weeks one to seven. This group was most successful in week six.

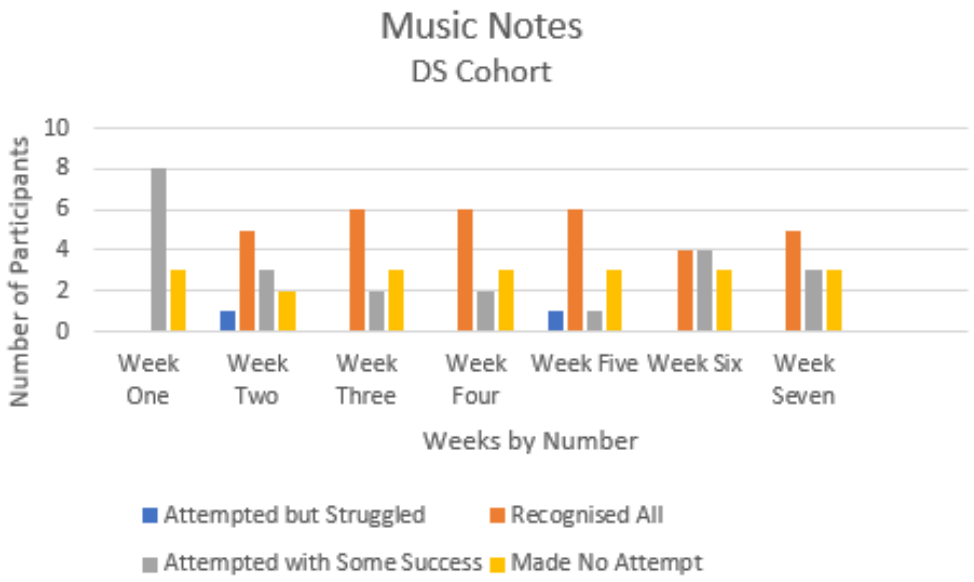


Figure 3.17. The number of children with DS who engaged with the music notes and the level to which they were successful in their recognition from weeks one to seven. This cohort was most successful in weeks 3-5 and then declined until week 7.

In summary, instrument recognition and the sequential songs were the most accessible components of the intervention. Figure 3.18 shows overwhelmingly that singing and actions were the preferred level of engagement with the sequential songs, with 19% engaging with actions only and 3% engaging with song.

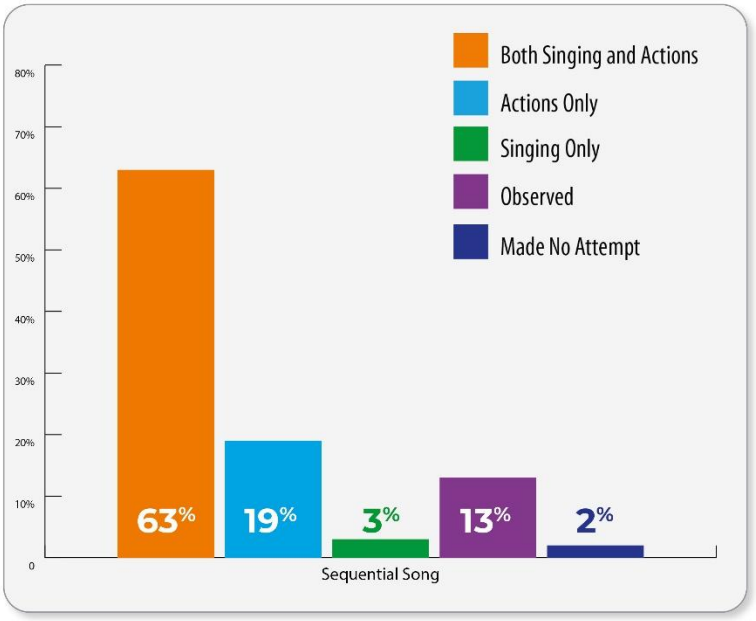


Figure 3.18. The percentage and level of engagement with the sequential song, including singing and actions, singing only, actions only, those who observed, and those who made no attempt. Singing with actions was by far the preferred level of engagement with the song.

Figure 3.19 shows that children had similar success with the instrument parade and the instrument recognition, with significantly less success in recognising the music

notation. However, 37% of participants attempted the music notes with some success. A comparable number of children made no attempt at the instrument parade and the music notes.

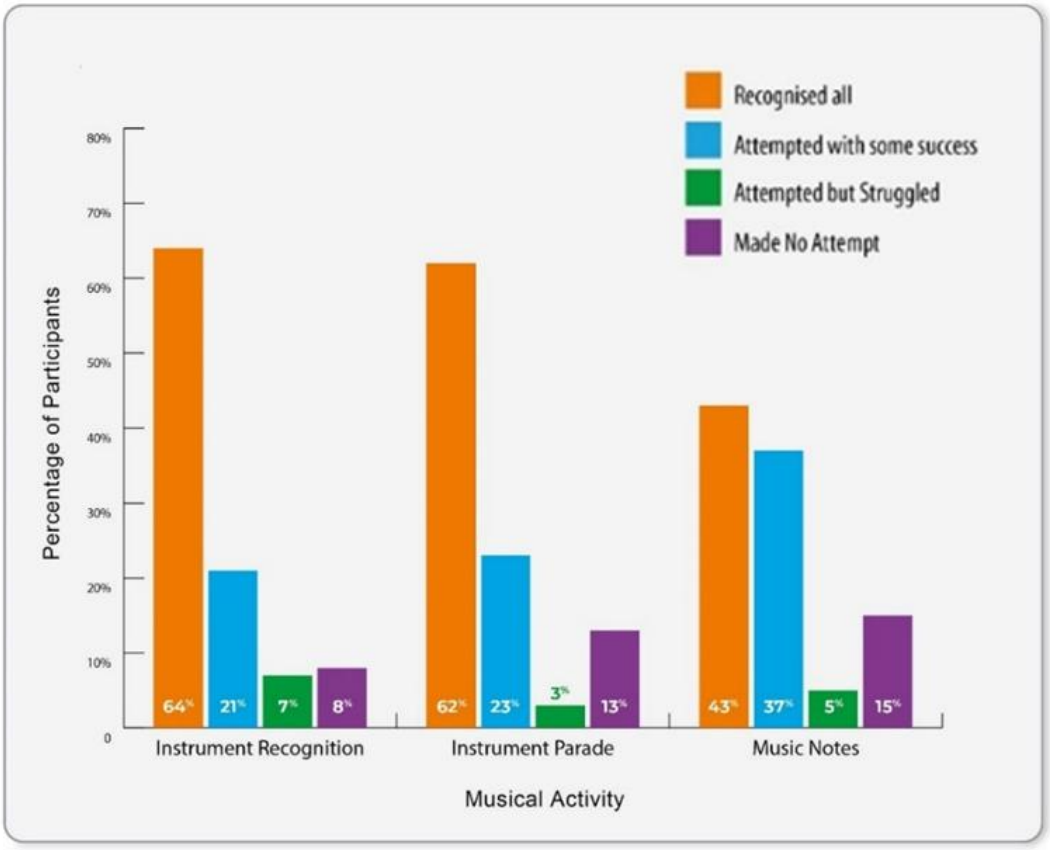


Figure 3.19. The percentage and level of engagement across both TD and DS cohorts to instrument recognition, the instrument parade, and the music notes. Children scored comparatively on the instrument recognition and instrument parade, with less success recognising the musical notes.

3.3.2 A Qualitative Parental Description of Your Child's Experience of Music

During the initial wave of the project, an additional questionnaire, 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention,' received ethical approval to capture both parents' and guardians' observations and thoughts on how their child experienced the intervention. However, ethical approval for this questionnaire was only granted in March 2022 after wave one was completed and wave two was nearing completion. Therefore, 15 participants were recruited after ethical approval was granted. Of the 15 guardians that completed the questionnaire,

6 were parents of children with DS, and 9 were parents of TD children. The questionnaire included closed and open-ended questions, as seen in Table 3.7.

- Were there aspects of the music intervention videos that your child most enjoyed? If so, please indicate which. Please elaborate on your answer.
- The online class was an important part of the intervention (0 being not at all important and 10 being extremely important). Briefly explain your answer.
- Any other comments about the online classes and weekly video?
- Did your child sing or hum any of the songs outside of music class? (0 being that you did not sing the songs at all outside of the class, with 10 being you sang all of the songs outside of the class). Please elaborate on your answer.
- Do you or another guardian use any of these songs at home with your child? (0 being that you do not sing any at all, with 10 being you sing them all of the time). Please elaborate on your answer
- Please tell us more about any developmental observations in your child.
- Please tell us anything else that you think is relevant about your child's response to the music intervention.

Table 3.7. The open-ended questions included in 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention' questionnaire.

When asked, 'Were there aspects of the music intervention videos that your child most enjoyed? ' 75% of the TD cohort preferred the instrument parade, 62.5% of the cohort enjoyed the song singing. Similarly, 83% of the DS cohort enjoyed the instrument parade, while a 100% of children with DS enjoyed the song singing. Only the DS cohort enjoyed the rhythm family clocks, and the music vocabulary activity scored the lowest among both cohorts (Table 3.8, Figure 3.20).

Activity	TD Cohort %	DS Cohort %
Instrument Parade	75	83
Song Singing	62.5	100
Actions	37.5	100
Rhythm Family	37.5	83
Rhythm Family Clocks	0	67
Music Vocabulary	37.5	50

Table 3.8. The results of question one, 'Were there aspects of the Music Intervention Videos that your Child Most Enjoyed?' from the Parental Qualitative Description of Your Child's Experience of the Music Intervention.

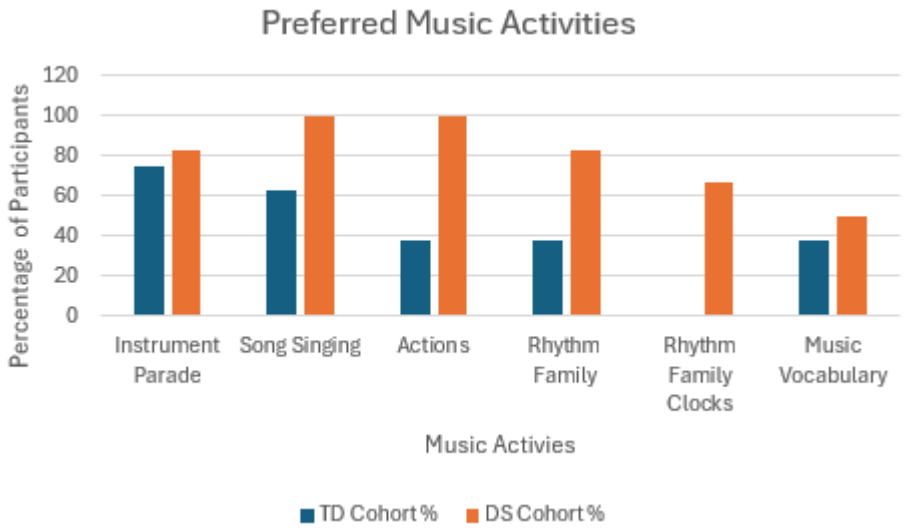


Figure 3.20. The responses from parents when asked to rate their child's preferred music activity.

When asked what they viewed as the optimum number of times to watch the video, both cohorts responded that they would watch it four times a week. Five times was the next preferred option in the DS cohort, while every day was the second preferred option in the TD group. The preferred duration of the videos was 15 minutes, as seen in Figure 3.21.

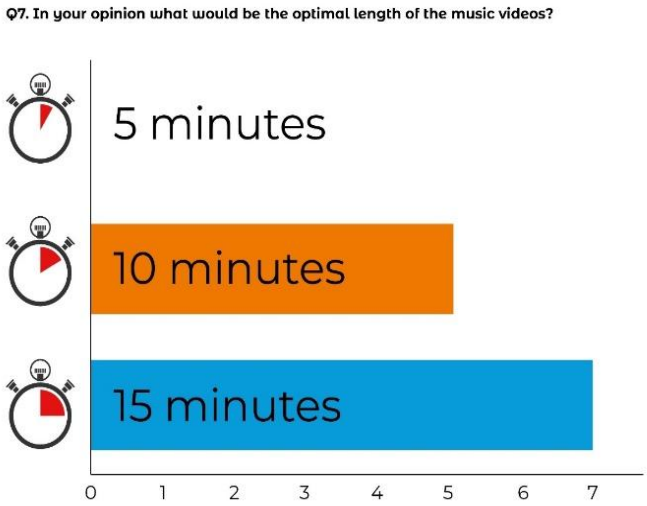


Figure 3.21. When parents and guardians were asked, 'In your opinion, what would be the optimal length of the music videos?' The optimum length of the online video was 15 minutes with 10 minutes being the next preferred option.

The parents/guardians of children with DS considered the online class more important, with 2/3 of the responses stating that it was very important (Likert Scale

point 10) and the remaining 1/3 identifying that it was important (Likert Scale point 9).

Parents were asked to reflect on the longer-term influence of the music intervention. They were specifically asked if their child sang any of the songs, with 0 being that they sang 'none of the songs' and 10 being they sang 'all the songs'. As can be seen in Figure 3.22, the TD cohort sang the songs more frequently outside of class time than the DS group.

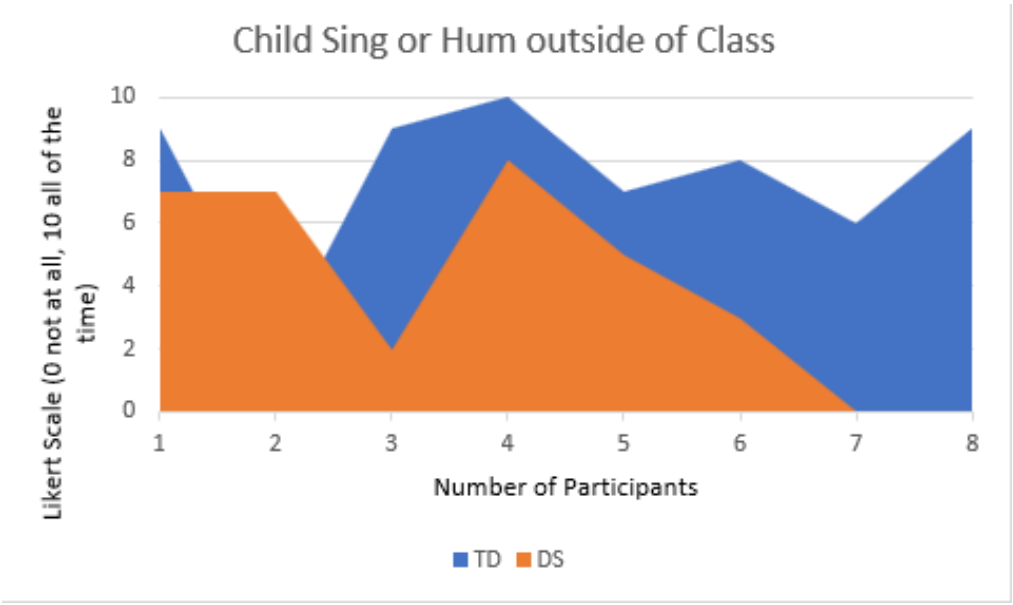


Figure 3.22. The number of responses to the question, 'Did your child sing or hum any of the songs outside of music class? ' (0 being that you did not sing the songs outside of the class, with 10 being that you sang all of the songs outside of the class). As is evident in the graph, the TD cohort sang the songs more frequently outside of class time than the DS group.

To assess the parental engagement with the music intervention songs, the parents were asked if they sang the songs with their children. Figure 3.23 shows that the parents of TD children sang more often outside of the intervention than the parents of children with DS.

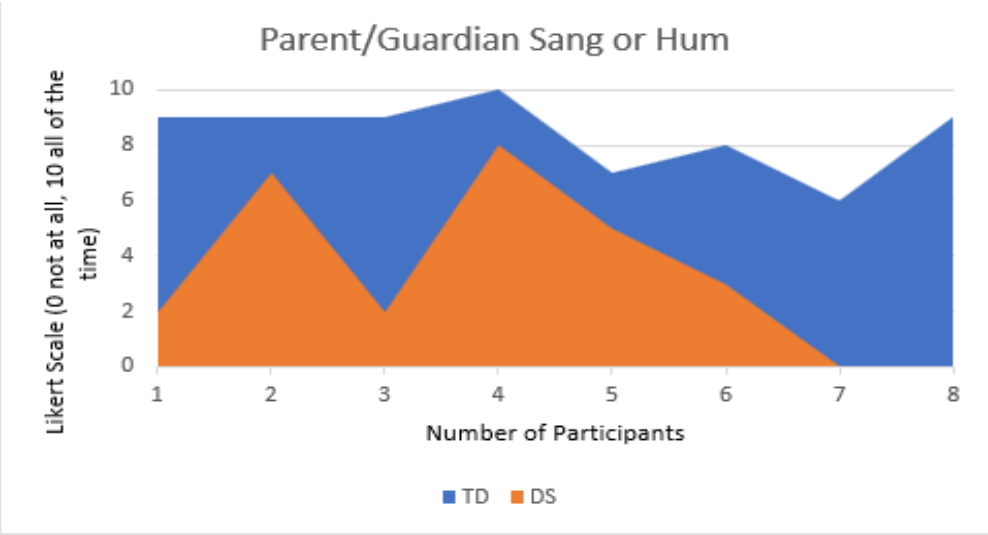


Figure 3.23. The number of responses to the question 'Did you sing or hum any of the songs outside of music class? (0 being that you did not sing the songs outside of the class, with 10 being that you sang all of the songs outside of the class). The parents of TD children sang the songs more often outside the class than those of children with DS.

When parents/guardians were asked if they noticed any developmental changes in their children due to engagement with the music intervention, there was a varied response. The scoring system was such that a score of 0 indicated that parents/guardians noticed 'no change', and a score of 10 indicated that they observed 'a lot' of developmental changes. Four participants noted no changes in their child's development, and six scored their observations above 5/10 on the Likert scale. Two parents showed a response above eight on the scale. Therefore, 11/15 (73%) noticed some developmental changes in their children because they engaged with the music intervention.

The parents/guardians were also asked if they had seen an improvement in any specific area of their child's development. Parents/guardians of children in both cohorts observed noticeable improvements in communication and cognitive skills, as seen in Figure 3.24.

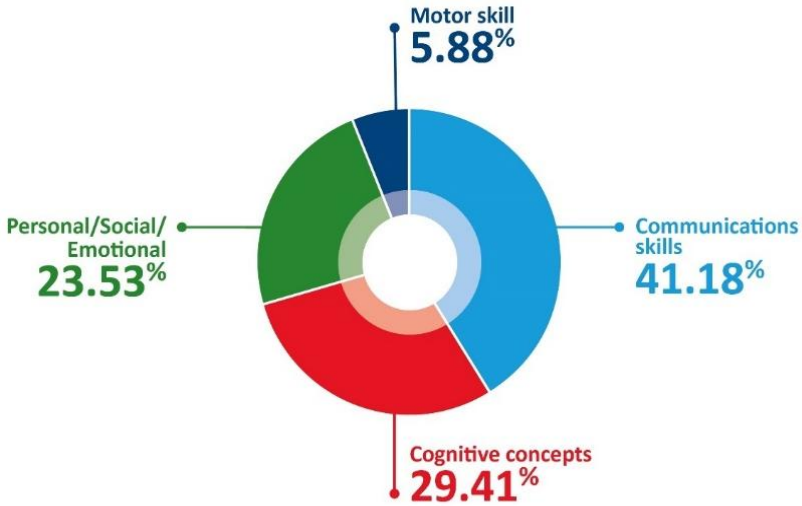


Figure 3.24. The percentage of recognised areas of improvement as identified by the parents. Communication was the most recognised area, with cognitive concepts being the second, personal/social/emotional third, and motor skills the least impacted area.

The open-ended questions allowed parents and guardians to elaborate on their selected answers and reflect on their observations. The answers were coded (a sample of which can be seen in Appendix C) and divided into the following categories and subcategories, as seen in Figure 3.25.

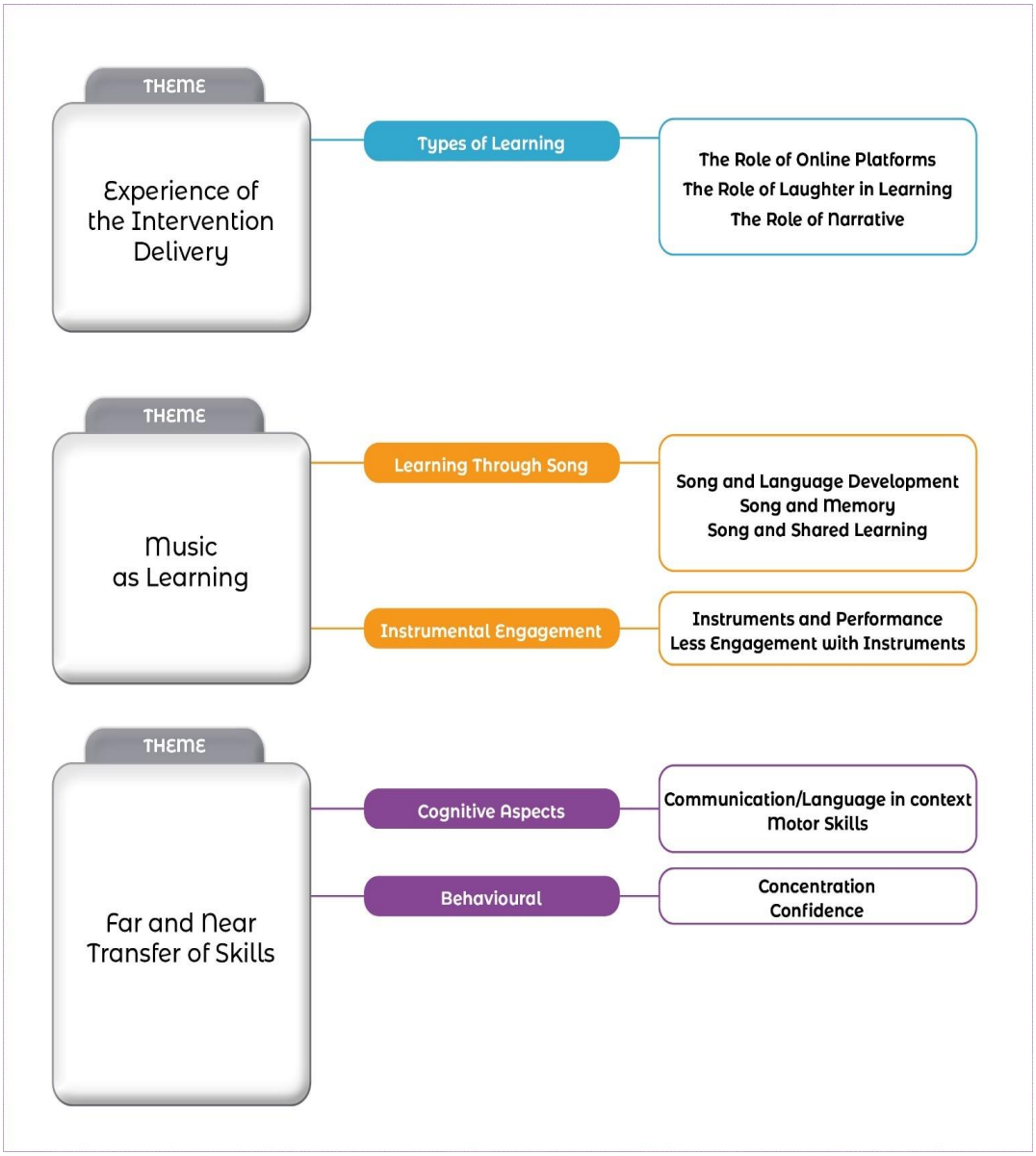


Figure 3.25. The content analysis of responses to the open-ended questions in 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention', Themes, Categories and Subcategories.

After analysing the qualitative piece from the survey, three specific themes emerged: **the experience of the intervention delivery**, **music and learning**, and **the far and near transfer of skills**. Each theme is subdivided into two categories with several subcategories.

3.3.3 Experience of the Intervention

One of the emergent themes from the qualitative data was the participants' **experience of the intervention**. The subcategories formed from this included contrasting forms and **methods of learning**. **The role of the online platform** and the

contrasting delivery methods received much attention from the participants' parents. There was an online class on Teams and pre-recorded videos on YouTube for the participants to look at, and as one parent noted, "seeing the contrast between these two forms of delivery was interesting." One noticed the positive effects of using online formats: "while digital technology and devices are criticised a lot, it was great to see how they can also positively impact learning." The ability to re-visit the YouTube classes at any time was noted as "we still go back and replay the music intervention videos on a Saturday morning when [their] sister is at her music lesson/on a rainy weekend/sick day from school." Another "still watches some YouTube clips and plays with the egg shaker and tambourine." Also, the accessibility of this format was noticed, as "she would recognise the songs and look to play them on YouTube" while another "watched them on YouTube himself." Others transferred the learning to another device, such as the Alexa.

There were positive and negative responses to engaging with the live online class as one "did not like the online class, probably because they are not used to Zoom." In contrast, others found the impact of the online class encouraged the singing of songs as they sang them on a "Saturday after attending the Zoom lesson."

The survey highlighted **the role of narrative in learning about music** and emphasised how it supported subsequent learning. As identified by one of the participant's parents, this was evident in relation to the 'rhythm family'. The stories enabled the child to connect with the material more meaningfully, which encouraged this child to "make up some stories about them during the week."

The emphasis on enjoyment and **laughter in learning** was an evident subcategory. This was noted by several parents who found the programme delivery entertaining, as their children "found Pádraig very funny and loved when he acted silly in the videos." Similarly, another participant echoed this, thinking the "enjoyment factor was great to see." This was reflected in both the instrument playing and song singing. One participant mentioned how their child "enjoyed using the instruments to perform" while other siblings "still sing and laugh about the songs together and correct each other if one gets a bit wrong."

3.3.4 Music as Learning

Under the theme of **music as Learning**, the category of **learning through song** is featured strongly, with several subcategories becoming evident. There was much referral to **song and language development, song and memory**, and **song and shared learning**. The links between **song and language development** included many references in the feedback shared. One parent said, “we all sang a lot of the songs, and the instruments were used lots in day-to-day life (as part of SLT, too!).” Another parent noted the impact of this musical language, stating, “carry over of the **learning** was noticed with later comments from the child using piano and forte.” This transfer of **learning** was also evident in other contexts, specifically in relation to certain songs. One participant found that “the 'High Low' song made a particularly significant impact! My son would start singing this if the words 'high' or 'low' were mentioned in a sentence!” It was noted that the songs not only added to children’s vocabulary, but one parent also noted that they “heard [their child] using the melodies and putting different lyrics to the songs.”

Songs and Memory were also identified as a subcategory of music **learning**. However, people’s engagement differed quite considerably. While one participant found that their child did not engage with the songs or singing “as it is not something he does anyway.” Others found that they did not sing them at all outside the intervention, with one saying they sang them “an odd occasion.” Another participant sang them “now and again”, while others said it “did not occur to them” to sing them, or another kept “meaning to but forgot.” One parent found that some “songs were more memorable than others.” In contrast, one participant used to hear her children “singing the songs all the time, and still do now and again,” and another participant “still sings them in the car.” Some sang songs depending on their memorability, such as the “Hello Everyone song in particular.” Another participant found that they were surprised and very impressed at their child’s ability to remember the names of the people in the rhythm family, *the tempi,* the instruments, and the music terms.

Songs as Shared Learning and the dissemination of the songs with siblings and family members were found during the intervention. One participant noted that the child was “singing the songs to her younger sister.” Another found that even

though the participant involved in the intervention did not sing them, “his younger sibling, who is 3, really loved them.” The **shared learning** was also evident in another parent’s experience as their child “wanted to **share the learning** and show off what they knew. They also had loads of chats with his sister about what they were **learning**. The two of them could be heard singing and dancing to the songs they learned along the way.” The impact of the song singing during the intervention on the entire family was an evident theme in the data, with many parents noting the influence on their household as “we sang them all of the time during the intervention.” This was echoed by another individual who stated, “that the songs became a part of the household, particularly for the duration of the intervention.” The songs were introduced by the child taking part in the intervention or “started by me as they were in my head”, then the “family became familiar with them,” or “they rubbed off on us eventually.”

3.3.5 Far and Near Transfer of Skills

When asked about the developmental changes the parents and guardians noticed post-intervention, the theme of **far and near transfer skills** emerged. This was then subsequently divided into **cognitive and behavioural aspects**. Changes in gross **motor skills** were evident as one participant found that their “son kept time to the music perfectly (shaking, clapping, stomping).” Noticeable changes were also found in fine **motor skills** by a participant who found that their “son learned to use the space bar to stop the video (to find an instrument/ pipe cleaner (music note) and restart. He has progressed to using the computer mouse!”

Language and communication in context were also recognised as a category under **cognitive** aspects. While it was “hard to tell what a natural progress was or due to the intervention,” many participants recognised the change in the use of **language**. One parent noticed “she made strides in her schoolwork, **communication** skills, listening and learning.” Another participant said, “their son learned and retained the instrument names and music terms (e.g. crescendo, largo, presto).” Others “were surprised and very impressed at [their child’s] ability to remember the names of the people in the rhythm family, the tempo, the instruments, the music terms.” One participant noticed their child “attempted to say the music vocabulary.” An additional participant noticed that their child used a lot of the musical **language** that she would

not have been familiar with before the intervention." This participant noted that their child "interestingly…used [these] only in a musical context and not outside." In contrast, one participant recognised that their child "used the musical **language** and outside of a musical environment. For example, if someone was being loud, he would say, 'please do not be *forte*'."

In the category of behavioural aspects, many participants noted confidence. For example, one parent found that their "son gained **confidenc**e learning a new language like music and that **confidence** made him more vocal." Another parent found that their son "really enjoyed the music, and he now will sit at the piano (which arrived at XMAS) and has the **confidence** to bang out a made-up tune. He would not have done that before." Another aspect that received emphasis was **concentration**, as one parent found the main change "would be **concentration**". Another parent felt that their son's "**concentration** levels improved slightly."

3.4 The Impact of the Intervention on Temporal Order Judgement and Short-Term Spatial Memory.

All cohorts were required to complete TOJ and Corsi Blocks tasks as part of the intervention. As seen in Table 3.9, only 15 out of 46 participants completed the pre- and post-measure, with a compliance rate of 32.6%. Of these 15 participants, 11 (73.3%) were TD children engaging with music, whereas 2 (13.3%) were in the TD control. No participants from the DS control group completed the task, while only 2 (13.3%) from the DS group engaged with music complied.

Group	Corsi Blocks Pre- N	Corsi Blocks Post- N	Temporal Order Judgement Task Pre- N	Temporal Order Judgement Task Post- N
TD No Music	**2 (8%)**	**2 (8%)**	**2 (8%)**	**2 (8%)**
TD Music	**11 (20%)**	**11 (20%)**	**11 (20%)**	**11 (20%)**
DS No Music	**0 (0%)**	**0 (0%)**	**0 (0%)**	**0 (0%)**
DS Music	**2 (8%)**	**2 (8%)**	**2 (8%)**	**2 (8%)**

Table 3.9. The number of participants in each group who engaged with the Corsi Blocks and Temporal Order Judgement Tasks, both pre- and post-intervention. The parenthesis represents the percentage of the group that completed the task.

The TOJ analyses were based on a Group 3 (TD No Music, TD Music, DS Music) x Time 2 (pre- and post) x SOA 7 (50 – 500 ms) mixed-design ANOVA, with Group as the between-subjects factor, Time and SOA as the within-subjects factors. Unimodal blocks (auditory- and vision-only) were analysed separately. There was no significant main effect of Group [$F(1,12) = 2.37$, $p > 0.05$]. There was also no main effect of Group [$F(2,12) = 2.6$, $p > 0.05$]. There was no significant interaction between these factors [$F(2,12) < 1$, n.s.]. It is important to note that these analyses are exploratory as the number of participants is insufficient for inferential statistics.

Auditory-only

There was no main effect of Group [$F(2,12) = 1.45$, $p = 0.273$]. There was no main effect of Time [$F(1,12) < 1$, *n.s.*] or SOA [$F(1,12) = 4.62$, $p = 0.053$]. Finally, there was a significant interaction between SOA and Group [$F(2,12) = 5.38$, $p = 0.022$]. As the SOA increased, the TD groups became more accurate, while the DS-music group decreased in accuracy as the SOA increased. There were no other significant interactions, as seen in Figure 3.26.

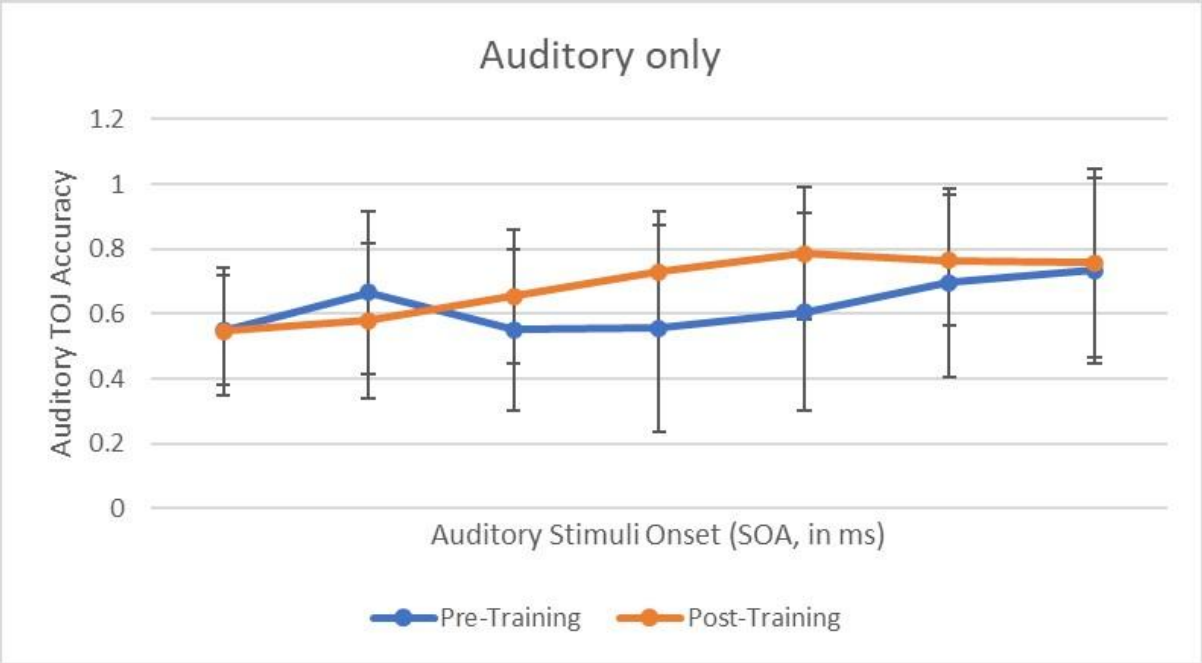


Figure 3.26. Both pre- and post-intervention auditory responses detailing auditory temporal order judgement accuracy and stimulus onset asynchrony.

Vision-only

There were no main effects of Group [$F(2,12) < 1$, *n.s.*], Time [F(1,12) = 2.59, p = 0.134] or SOA [F(1,12) = 2.96, p = 0.111]. Also, there were no significant interactions (all *p*s > .05).

Multisensory

The multisensory condition included both audio and visual modalities; the visual-lead condition was when the flash was seen first, and the auditory condition was where the beep was heard first. We ran four groups; Time was pre and post with 7 SOA (+/-50ms - +/-500 ms). The auditory lead had no main effect on Group [$F(2,12) = 0.161$, *n.s.*]. There was an effect of Time [$F(1,12) = 8.86$, p = 0.12] and SOA [$F(1,12) = 9.91$, $p = 0.008$]. There was no significant interaction in relation to SOA and Group [F(2,12) = 1.81, p = .20]. There was also no significant three-way interaction between [$F(2,12) = 2.56$, $p = 0.12$].

In the visual-lead condition, we assessed the main effect in the group collapsed over time. The vision-lead responses had no significant main effect on Group [F(2, 12) = 4.42, p = 0.35. There was also no significant main effect of Time [F(1,12) = 1.96, p = 0.186], but there was a significant main effect of SOA [F(1,12) =

7.08, p = 0.021]. There was no significant interaction between SOA and Group [F(2,12) < 1, *n.s.*] or in Time, SOA and Group [F(2,12) = 1.16, p = 0.344]. Therefore, there was only an improvement in the SOA in the visual lead component (Figure 3.27).

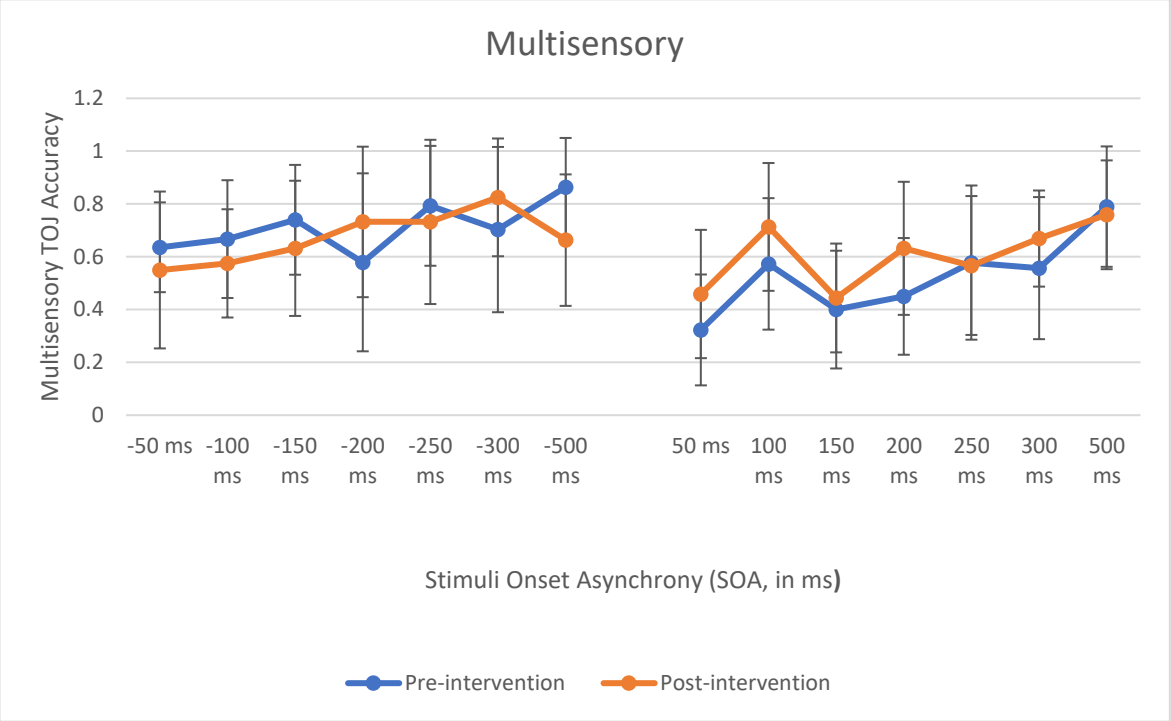


Figure 3.27. Both pre- and post-intervention, multisensory responses detailing multisensory temporal order judgement accuracy and stimuli onset asynchrony.

Corsi Blocks

There were 3 Groups (TD No Music, TD Music, DS Music) x 2 Time (pre-post-Corsi score) design in the Corsi task. An ANOVA was conducted, and there was no main effect of Group [*F*(1, 12) = 2.374, p = .149] or Time [*F*(1,12) = 2.559, p =.119]. There was also no significant interaction between these two factors [*F*(2,12) = .901, p = .432]. These analyses should be taken cautiously as the number of participants is insufficient.

3.5. The Impact of the Intervention on Paediatric Quality of Life (PedsQL)

The parents/guardians completed the PedsQL form relevant to their child's age (2–4 years, 5–7 years, or 8–12 years) before and after the intervention across all TD and DS groups, both engaging and not engaging with music (Vicari et al., 2001). Only participants who completed the forms both pre- and post-intervention were included in the results. Out of 46 participants, 22 were compliant (47.8%); 12 completed the

form for aged 8–12 years, and 10 completed the form for 5–7 years. No participants filled in the 2–4 years form. The number of respondents for each age category is presented in Table 3.10.

Age Group	TD Engaging with music	TD Not Engaging	DS Engaging	DS Not Engaging
2–4 years	0	0	0	0
5–7 years	8	2	0	0
8–12 years	2	0	8	2
Total	10	2	8	2

Table 3.10. The number of respondents who completed the Paediatric Quality of Life Form with children aged 2-4, 5–7 or 8–12 years.

Table 3.11 contains the cumulative results from age profiles and functions across all four groups. The higher scores indicate a better health-related quality of life (HRQOL).

Parent Report	DS Engaging		DS Control		TD Engaging		TD Control	
Core Domains	**Mean**	**SD**	**Mean**	**SD**	**Mean**	**SD**	**Mean**	**SD**
Physical Functioning Before Intervention	54.2	9.75	60.9	14.5	81.56	12	98.4	4.1
Physical Functioning After Intervention	54.37	15	57.81	12.40	80.6	11.2	75	10.82
Emotional Functioning Before Intervention	70.59	9.37	65	14.6	62.5	3.53	65	9.35
Emotional Functioning After the Intervention	77	7.4	65	9.35	68.5	7.17	60	9.35
Social Functioning Before Intervention	43.75	21.89	45	23.1	81.5	5.14	85	12.2
Social Functioning After Intervention	62	15.3	47.5	24.24	82.5	6.89	80	6.1
School/Work Functioning Before Intervention	61.25	11.27	52.5	18.3	82.5	6.89	87.5	11.18
School/Work Functioning After Intervention	67	12.88	52.5	24.23	83.5	6.2	85	5

Table 3.11. The mean and standard deviation results from the Paediatric Quality of Life Inventory across all functions and all TD and DS groups.

The number of participants is too low to consider statistical significance. The results are presented in Figures 3.28, 3.29, 3.30, and 3.31.

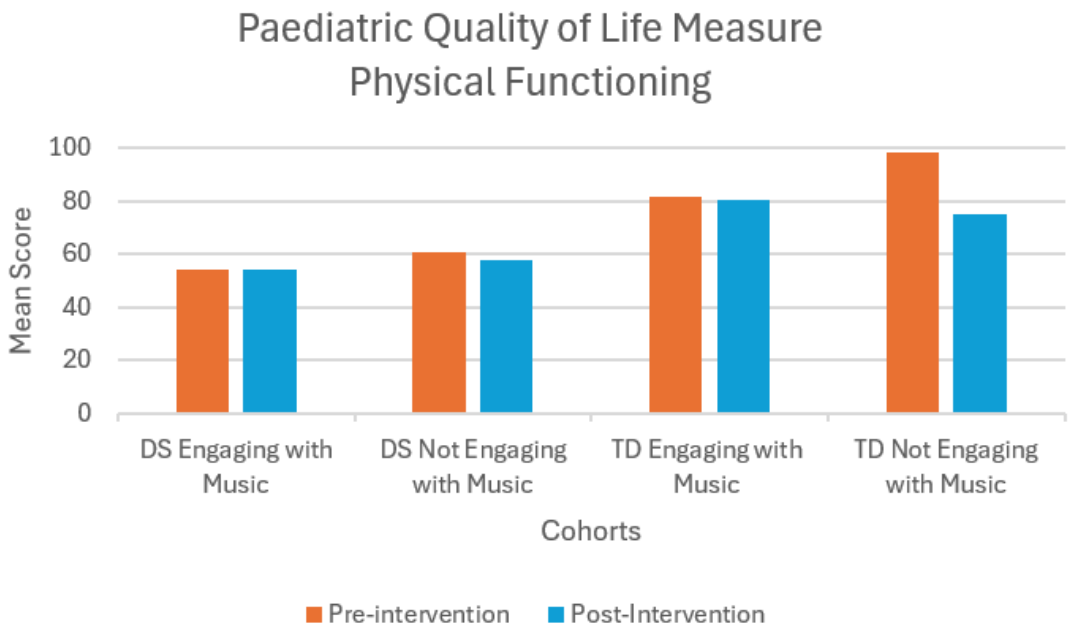


Figure 3.28. The results of the Physical Functioning of all cohorts in the Paediatric Quality of Life measure both pre- and post-intervention

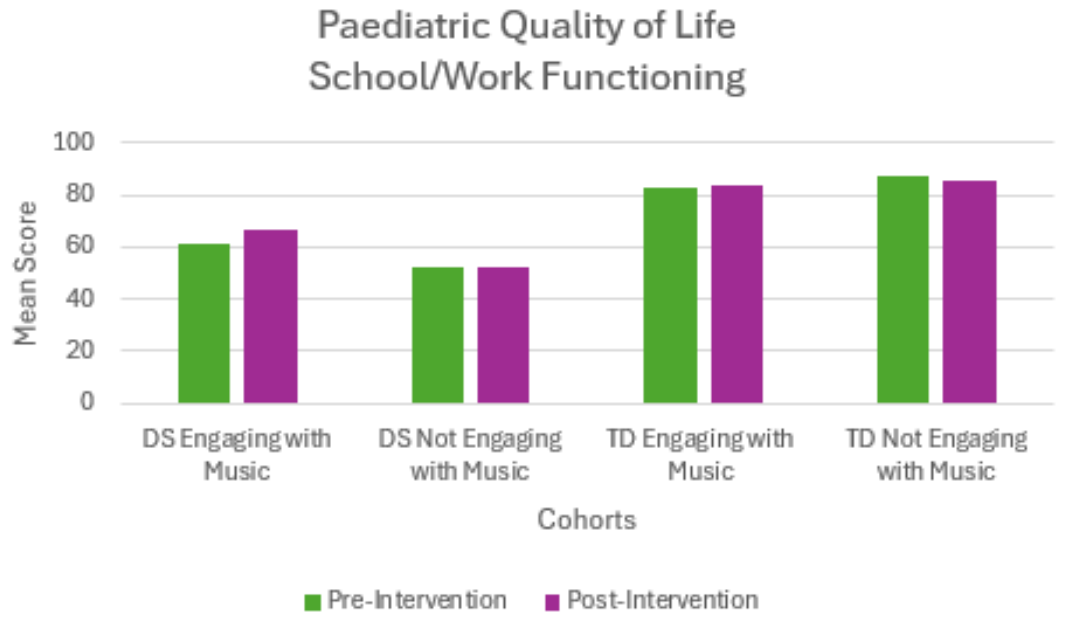


Figure 3.29. The results of the School/Work Functioning of all cohorts in the Paediatric Quality of Life measure both pre- and post-intervention.

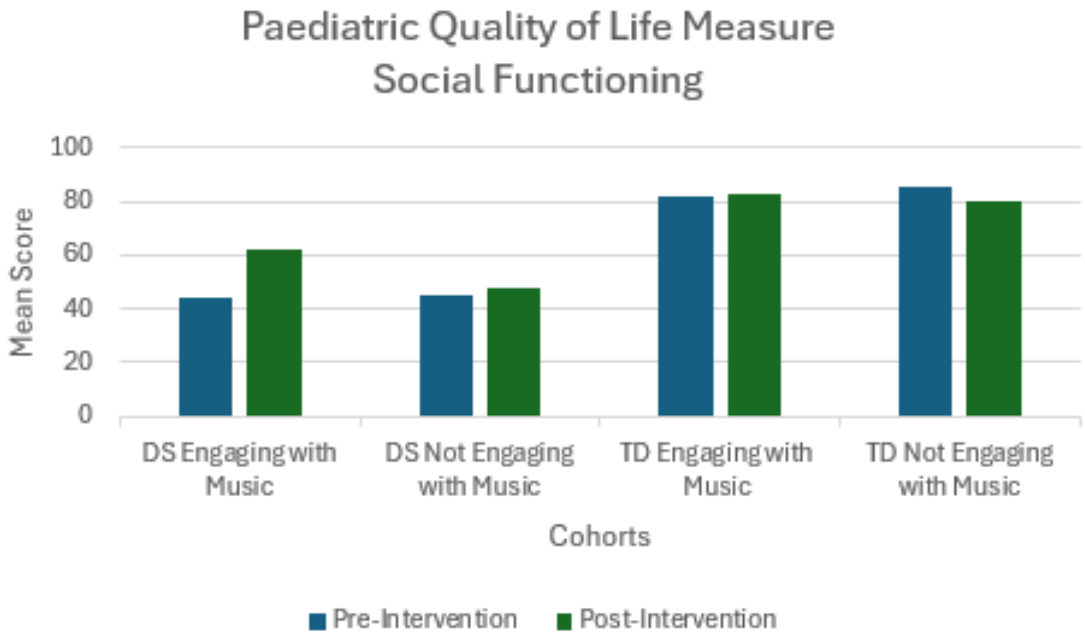


Figure 3.30. The results of the Social Functioning of all cohorts in the Paediatric Quality of Life measure both pre- and post-intervention.

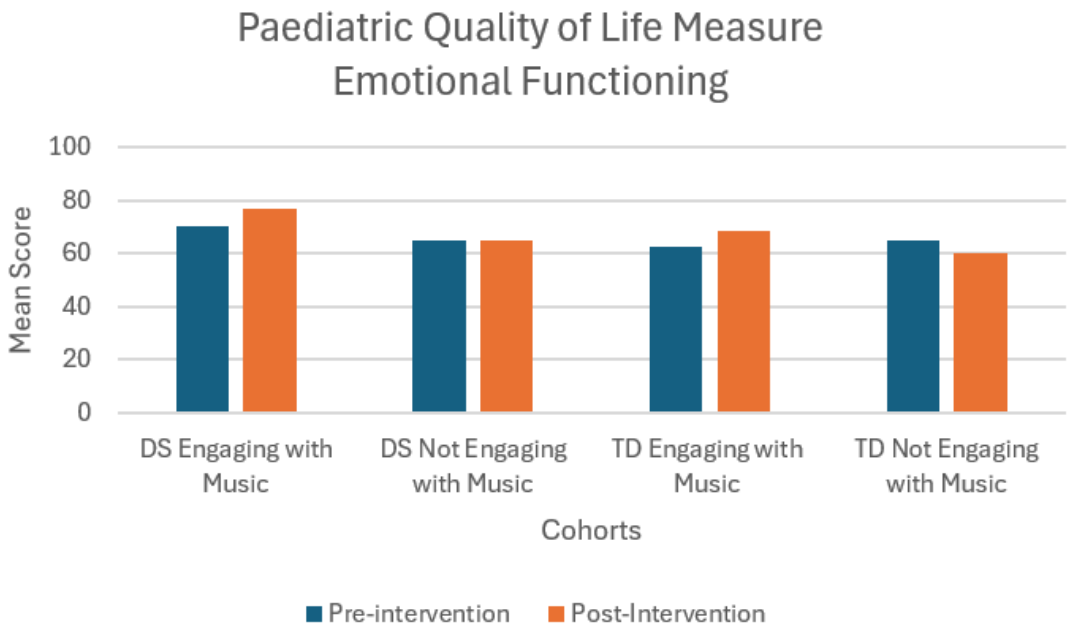


Figure 3.31. The results of the Emotional Functioning of all cohorts in the Paediatric Quality of Life measure both pre- and post-intervention.

3.6 The Impact of the Music Intervention on Cortisol and Oxytocin Concentrations.

Hair and saliva samples were collected from 46 participants before and after the intervention. There were 46 hair samples collected before and after the intervention, with 92 samples collected in total, two from each participant. Three saliva samples

were collected from each participant, both pre -and post-intervention, resulting in a total of 272 saliva samples collected. However, this was not sufficient for statistical power to measure cortisol concentration. Therefore, the samples were appropriately stored to be analysed at a later date.

Discussion

The aim of the study was to assess temporal perception, visual memory and alterations to cortisol and oxytocin before and after a music intervention in TD children compared to children with DS. At the end of the study, we observed the following.

- There are challenges in recruiting adequate numbers from the DS cohort for such studies.
- There is limited availability of suitable music education programmes for children with DS.
- There were differing levels of engagement in the online platforms between the cohorts.
- Children enjoyed the intervention, particularly song singing and actions.
- There were challenges with compliance with the TOJ and Corsi Blocks test, which did not then show any significant difference pre- and post-intervention.
- There are some indicators to show that engaging with a music intervention can impact a child's quality of life.
- An increased number of participants is required to sufficiently power the analysis of biological samples and thus the impact of the music intervention on cortisol and oxytocin concentrations was not determined in this study

A detailed examination of these observations will form the discussion that follows.

4.1 Recruitment

The initial recruitment profile of the parents/guardians engaging with the MINdDS programme was predominantly female. This is in accordance with the literature (Smith, 2008), as it was found that 75.6% of parents who filled out the questionnaires were female, with only 13.5% of these being homemakers. Therefore, the responses were female-dominated despite most of them working outside the home. All parents and guardians participating in the study were in the 30–49 years and 50+ age categories. Those who were in the latter age category had children with DS, which coincides with existing research that there is a link between maternal age and increased chances of having a child with DS (Savva, Morris et al., 2006).

As has been reflected in the literature, recruiting significant numbers of children with DS for studies in music can be challenging (Gemma, Pablo et al., 2020). Similarly, the total number of participants involved in MINdDS after withdrawal was

46. There were 26 TD children (m=11 and f=15) and 20 children with DS engaged in the study (m=12 and f=8). The ratio of boys with DS in this research project coincides with a survey by Verma, who found a higher occurrence of DS in males than females (Verma, 1987). Moreover, similar to that reported in a review by Yang & Conners et al. (2014), there was a considerable disparity in the age range of the children when MA-matched. For example, while the average CA of those with DS who participated in the study was 10.5 years in the music intervention group and 9.7 years in the control group, when MA-matched, the music group had an average age of 4.3 years, while the control group had an average of 3.8 years. The disparity between the CA range of the children with their MA equivalent is substantial. It shows the importance of using a measure such as PPVT-4 to establish the MA of children with DS before an intervention. The disparity in age profiles between groups and within groups meant that the material in the music intervention was more accessible and appropriate to some and not to others, which impacted attrition. This was reflected in the 9 participants who withdrew from the music intervention groups: 7 from the TD group and 2 from the DS group. Seven of the eleven males recruited in the TD group withdrew from the music intervention. This is in keeping with the literature that has found a lower percentage of males tend to engage with music programmes than females (McPherson and O'Neill, 2010). It could also be reflective that the content needed adjusting and was more suitable for younger children.

4.2 Limited Availability of Accessible Music Programmes

MINdDS highlighted the lack of engagement of all cohorts in music programmes; only 22.8% of the children had previously engaged in music. The availability and uptake of music as an extracurricular activity for children is still limited (Conaghan, 2021). Parents and guardians stated a number of reasons for this lack of engagement, including an over-commitment to existing activities, the age of the child, and children not being interested. However, the prevailing response was the lack of availability and, more importantly, accessible music programmes for their child. This limited availability is particularly evident for children with DS, as one parent noted that while there was more availability for her other TD children, there were no classes "specifically for children with DS; hence [there was] little opportunity to learn." It was

also noted by parents that they felt that tutors and teachers did not have the relevant expertise to teach children with ID, which led to 'frustration' for the children and parents. The need for teachers to upskill and gain an insight into how to adopt a more inclusive practice has been acknowledged in the literature (Gyasi, et al., 2020). The void in the ability to facilitate creative practice for people with ID has also been emphasised, as well as the noticeable gap in providing music education outside the realm of music therapy for this cohort (Gemma, Pablo et al., 2020; Després et al., 2023). Therefore, this intervention created a platform to introduce the children to music fundamentals and musical terms in a way that was accessible to children of all abilities.

4.3 Online Delivery: Levels of Engagement

During the MINdDS project, the participants were required to engage with a pre-recorded YouTube video five times a week (video length varied from 15 to 30 minutes) and one live class online. Both the class and the music videos were delivered through online platforms, and as one parent said, "seeing the contrast between these two forms of delivery was interesting." as the compliance and level of engagement differed considerably. There was a compliance rate of over 70% with the YouTube videos. The YouTube videos could be viewed at a participant's convenience at whatever time in the day that suited their family routine. The participants and their parents enjoyed the flexibility of this format, as well as the ability to re-visit the YouTube classes at any time. One parent noted that "we still go back and replay the music intervention videos on a Saturday morning when [their] sister is at her music lesson/on a rainy weekend/sick day from school." However, as acknowledged in the daily diaries, there were evident disparities between both groups regarding the amount of time they engaged with the YouTube videos over the timespan of the intervention. By week four, 85% of the TD group engaged with the video for 15 minutes. The remaining 15% watched the video for 10 minutes. By week six the number of TD participants watching the video for 15 minutes increased to 92%. In week seven, there was a slight decrease experienced in the TD group, with 78% engaging for 15 minutes from week seven. This could suggest that the TD group require a different level of engagement or intervention dosage than the DS group.

In contrast, while most children with DS engaged with the video for 15 minutes, there were more varied engagement levels, perhaps reflecting the differing abilities in this cohort (Karmiloff-Smith, 2016). For example, in week three, 55% of the DS group engaged with the video for 15 minutes, while 9% engaged for 10 minutes, 18% of the group engaged for 5 minutes, and 18% did not engage at all. However, by week seven, 91% of the DS group engaged for the full 15 minutes. This could indicate that the DS group required more time to increase their familiarity with the video, achieved through structure in their routine and repetition (Stein, 2001).

While everyone could engage with the YouTube videos without transitional issues, the success of the live class was divided. The live class comprised of a music practitioner teaching the class in real-time and at a specific time on a Saturday. This involved the practitioner leading the class, teaching concepts, and asking questions for 40–60 minutes. However, while the live class had an inherent interactive component, attendance at the class varied considerably from week to week. Mostly, this variation was because of other commitments on a Saturday, that families were away, or that some parents felt their child did not engage with the online class, as they "did not like the online class, probably because they are not used to Zoom." Each wave saw a decrease in attendance at the live class for the duration of the intervention, particularly in week six in both waves, with only 60% attending in week six wave one and 55% in week six wave two. No total attendance was recorded in wave two. The latter wave was less well attended, indicating that perhaps the fewer people who engaged with the programme could have diluted the sense of a group dynamic, leaving people less willing to be part of a small group. However, as one parent stated that "while digital technology and devices are criticised a lot, it was great to see how they can also positively impact learning."

4.4 Experiential Learning and Enjoyment of the Intervention

Song singing and the instrument parade were the preferred activities during the intervention. The popularity of song singing among children has been previously supported by a study carried out by Koutsoupidou (Koutsoupidou, 2020). Similarly, the MINdDS study noted that the songs were sung with the child by over 60% of the parents or a guardian 50% of the time. The children also sang with their siblings during

the intervention. The selected song types used in MINdDS were sequential songs. Sequential songs are songs that follow a certain pattern of repeated instructions. They often start with a smaller number of instructions which increase throughout the song. The songs selected had an upbeat tempo and limited melodic range to increase vocal accessibility and were accompanied by actions to encourage participation. 'Zippidy Zap' by Judy Cranson uses actions and instruments, while 'Music Music' released by Tumble Tots requires actions using lollipop sticks. Such songs were positioned at the start and the conclusion of the class to form a structural element. They contained the repetition that is required to optimise learning for all children, but specifically for those with DS (Stein, 2011). The impact of repetition in song singing has been acknowledged by Frizelle and McMullan, who found that increased exposures and higher dosages of song singing have the most significant impact on the speech development of children with DS (Barker, 1999; Frizelle, McMullan et al., 2024).

As seen in Table 4.1 such sequential songs reinforce procedural and episodic learning and memory via the cerebellum, HPC, and the PFC, which are areas of the brain that are impacted in children with DS (Sylvester, 1983; Guidi, Bonasoni et al., 2008; Volpe, 2009; Sathyanesan, Zhou et al., 2019). Emotional regulation and social bonding are mediated via the striatum, amygdala, and hypothalamus. While there are aberrations in the striatum and hypothalamus, the amygdala is unaffected in children with DS (Castells et al., 1996; Aylward et al., 1999).

Brain Function	Musical Method	Brain Region	Measures
Learning and Memory	Sequential Songs Songs with instructions and movement Synchronisation Instrument Parade	Cerebellum Hippocampus Prefrontal Cortex	Temporal Order Judgement Task Corsi Blocks Test PedsQl Quality of Life Inventory
Sensory-Motor Skills	Rhythm Family Improvisation Instrument Playing Group playing Beat Synchronisation	Motor Cortex Cerebellum Parietal Lobe	Temporal Order Judgement Task Corsi Blocks Test
Emotion Regulation	Music-Evoked Emotions through song	Striatum Amygdala Hippocampus Hypothalamus	Cortisol PedsQl Quality of Life Inventory
Social Bonding	Group Singing Group Performance	Striatum Amygdala Hippocampus Hypothalamus	Oxytocin PedsQl Quality of Life Inventory
Executive Function	Musical Clocks Rhythm Family	Prefrontal Cortex	Temporal Order Judgement Task Corsi Blocks Test PedsQl Quality of Life Inventory

Table 4.1. A summary of the related brain functions, musical methods, brain region and measures.

During the intervention, the TD cohort preferred singing and actions, with the level of engagement peaking in weeks six and seven, as 92% of the group engaged. The DS group similarly engaged with singing and actions; however, while in weeks one and two, the cohort engaged with both, the preference for observation and actions only increased from week two onwards. By week seven, there was almost an even divide between those who observed (27.2%), those who did the actions and singing (36.3%) and those who did actions only (36.3%). The reason why increased numbers either observed or engaged with the song through actions could be because some participants found it challenging to vocalise or verbalise the song due to language difficulties that are prevalent in this cohort (Filipe, 2022).

The instrument parade was also one of the preferred activities during the intervention and was related to another activity, instrument recognition. Both cohorts performed similarly in the instrument recognition as this required short-term memory only. In contrast, the instrument parade was devised to engage mainly working memory and visuospatial skills. The children were presented with an increasing number of instruments from week to week in the same order. It was structured according to Cowan's theory that the human short-term memory can retain 4 objects plus or minus 2 (Cowan, 2010). Therefore, there was a minimum number of two and a maximum number of six instruments presented to the children at any one time. The participants had to remember the order in which the instruments were shown and recognise the one that was missing. In the TD cohort, there was a steady improvement in the ability to recognise the instruments in the parade, with maximum engagement found in week six when 92% of the cohort recognised all. However, there was a decline in this engagement to 71% in week seven, perhaps suggesting that once the TD cohort recognised all the instruments their interest began to dissipate.

The group of children with DS engaging with music showed a contrasting learning trajectory with the instrument parade. In the initial week of the intervention, 36.3% of the group attempted with limited success in comparison to 45% that recognised all. The remaining participants either made no attempt or attempted and struggled. However, aside from weeks three and five, which saw an increase in those making no attempt, the DS cohort increased their ability to recognise the instruments, particularly in weeks six and seven, with 54% being able to recognise all. These results indicate that the DS cohort found this more challenging than the TD group. While their perirhinal cortex, which is responsible for object recognition, is intact, the DS cohort struggled more with the instrument parade as this relied on visuospatial working memory and short-term memory (Clark, Fernandez et al., 2017). This was similarly encountered in a study by Clark and Fernandez and consistent with the review by Yang et al., which states that while visuospatial working memory is considered a strength in this cohort, it is a relative strength compared to other abilities (Yang & Conners et al., 2014; Clark, Fernandez et al., 2017).

Finally, recognising the music notes was the most challenging for both cohorts in this intervention, with a success rate of 43% across both groups being able to recognise all of the notes. In week one, only 35.7% of the TD cohort successfully recognised the music notes, with 50% attempting with some success. Their success rate dramatically increased in week six, with 85% recognising all of the notes. As seen in previous activities, there is a dramatic decrease in the engagement of the TD cohort in week seven. Again, this points to perhaps a level of complacency and disinterest once the learning of the notes was achieved. The DS cohort showed a contrasting learning profile. In week one, 72% of the group attempted with some success, whereas the remaining 28% made no attempt. However, with this activity, the DS cohort's most success was achieved in weeks three to five, with a decrease in success rate for weeks six and seven. The number of those who made no attempt was consistent throughout the intervention, exposing the differences in ability and learning trajectory in this cohort (Karmiloff-Smith et al., 2016.

The experience of the intervention exposed parents and children to different forms of learning, in which the role of musical play was prioritised to increase the accessibility and enjoyment of the material (Marsh & Young, 2006). This was noted by parents who said, “the enjoyment factor was great to see.” This enjoyment was reiterated in several responses by parents and found in numerous subcategories as it “would be [the] main reason to encourage [their children’s] interests.” This was similarly found in the study by Koutsoupidou, who asked practitioners who work with children about what musical play means in their learning contexts; the researcher found that ‘play with music’ and ‘enjoyment’ were the most popular responses (Koutsoupidou, 2020). This musical play was also fostered through the effective use of narrative, where interactive stories about the rhythm family made the content more relatable. The stories enabled the children to connect with the material more meaningfully, which encouraged this child to “make up some stories about them during the week.” Using narrative and storytelling to enhance engagement with learning has been noted in literature across disciplines (Prins et al., 2017).

The impact of the association between music and fun in learning can often be undervalued. However, enjoyment is strongly partnered with successful learning (Hernik & Jaworska, 2018). If children are happy in themselves, oxytocin and

serotonin are released (Dsouza et al., 2020), optimising their learning capabilities (Carr & O'Mahony, 2019). In the words of one parent, "it is next to impossible not to engage with music in some way", and therefore, this makes it an accessible medium to children of all abilities that could give further insight into the developmental trajectories of neurodiverse populations.

4.5 The Impact of the Intervention on Cognitive Measures

The Corsi Blocks and TOJ tests were conducted to test short-term spatial memory and temporal perception, respectively. Both tests have been used with cohorts of children with DS previously (Yang & Conners et al., 2014). However, compliance with this measure was challenging, and many complained about the duration and complexity of the test. While the TD groups improved as the time-space increased in the unimodal auditory component, the DS cohort disimproved as time increased and hence became less accurate. In contrast, participants improved in accuracy in the visual lead component of the task. This confirms the higher functionality of visuospatial response in children with DS (Yang & Conners et al. 2014), while the participants' auditory response is more impaired. There was a minor increase by the DS cohort in the Corsi Blocks test from pre- to post-intervention, again confirming that visuospatial memory is a relative strength in this cohort (Yang & Conners et al. 2014). However, a conclusive effect could not be reported as the numbers were limited.

4.6 The Impact of the Intervention on Quality of Life

The PedsQL questionnaire was carried out before and after the intervention across each group (Vicari et al., 2001). While numbers were limited, the questionnaire exposed the main differences in the functioning ability, specifically in 'physical functioning', 'emotional functioning', 'social functioning', and 'school/work functioning', between the DS and TD cohorts. The DS cohort scored significantly less on the physical and school/work functioning (Schworer et al., 2021). However, there was a slight increase in school/work functioning after the music intervention in the DS cohort. Perhaps indicating that if the intervention period was longer, it may help increase these children's performance at work or school (Dumont, 2017).

While children with DS are considered to be very sociable, in comparison to the TD cohort, this group scored considerably less. This reaffirms the work of a previous study that found the language and communication difficulties encountered by people with DS impact their social abilities (Schworer et al., 2021). However, there was a marked increase in the social functioning of those with DS who engaged with music when their pre- and post-scores were compared. The impact of music on social interactions is being continuously researched with both TD and neurodiverse populations, as it is believed to heighten social communications (Tarr et al., 2014; Nummenmoa, 2021).

Both the TD and DS cohorts scored comparatively on emotional functioning. While there was an increase in the emotional function of both cohorts that received music, there was a marked decline in the TD cohort who did not engage with music. This is in keeping with the literature, which has included a number of studies that have explored the impact of music on emotional regulation (Peters et al., 2023). It must be noted that these findings should be interpreted cautiously as they reflect the limited number of people involved in the group that answered the questionnaire.

Related impacts on quality-of-life functions were also found in the responses to 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention' questionnaire, where 73% of parents noted some developmental changes. Under half (41.18%) noticed changes in communication, with other cognitive skills scoring second, while 29.41% noticed no changes. There were 23.53% of parents who noticed changes in social skills, with few observing changes in motor development. In the systematic review that examined the role of music in the lives of children with DS by Gemma, Pablo et al. (2020), cognitive skills and communication were the least researched within the literature on music and people with DS. Therefore, there is a potential disconnect between areas believed to be impacted mainly by music and those actually affected. The developmental changes in communication could also have impacted the perceived improvement in the social functioning scored in the PedsQL. As children with DS experience delays in speech, using music as a mode of speech and language therapy (SLT) could have potential benefits that could impact other areas (Barker, 1999; Knight, 2017; Frizelle, McMullan et al., 2024). This was recognised by one parent who said we all sang a lot of the

songs, and the instruments were used lots in day-to-day life (as part of SLT, too)." This was also found in the aforementioned study by Frizelle et al. in an intervention that explored the impact of song and sign (Lámh) on speech development in children with DS. This study found that children with DS who had a basic language level mainly benefited from a higher dose frequency through song and sign (Frizelle & McMullan et al., 2024). Additionally, one of the songs in MINdDS, There's a Beat in My Feet' by Teresa Jennings, requirauditores the children to use their kazoos. A kazoo is a plastic instrument a child holds in their mouth, which can help strengthen the orbicularis oris muscles required for speech (Guy & Neve, 2005; Jain, Prachi & Rathee, 2020). Therefore, there are substantial research avenues to further examine the impact of song on speech development.

4.7 The Impact of the Intervention on Cortisol and Oxytocin Concentrations

A number of 120 participants is required to sufficiently power the analysis of biological samples, and thus, the impact of the music intervention on cortisol and oxytocin concentrations was not determined in this study.

Limitations

While there were some interesting findings, there were, however, elements that restricted the project. Compared to other studies (Gomez-Scarpetta et al., 2012; Camps et al., 2017; Gemma, Pablo et al., 2020; Celia et al., 2021), the number of children with DS involved in the MINdDS project was considerably higher. However, the numbers were still insufficient and impacted data analysis. The ability to recruit additional participants was hugely impacted by the COVID-19 pandemic as parents found it challenging to commit and engage with the project in the circumstances. The subdivision of the smaller-than-anticipated participant numbers into four groups and the withdrawal of nine participants from the study impacted the statistical analysis. It also significantly affected the processing of the biological samples. Therefore, the statistical analysis was underpowered at baseline. While collecting hair samples was accessible to all parents, collecting saliva from the DS cohort was more difficult. Many children have additional sensory challenges, so holding the cotton bud in their mouths for any time was incredibly difficult. This, therefore, impacted the amount of saliva that was collected.

COVID-19 additionally affected the delivery of the study; while initially designed to be carried out in person, the pandemic forced the research team to redesign the study, moving every component online. The music class was moved to the online platform of Microsoft Teams, and as seen in the results, there was only total attendance recorded in week one of the initial wave, with no total attendance in wave two. Some of the measures, including the PPVT-4 and the TOJ and Corsi Blocks tasks, were also administered online, which made it more challenging for some to engage. The project involved several measures and required considerable commitment from those involved in the study. Therefore, the inability to meet with people in person during the pandemic contributed massively to the lack of compliance with a lot of the measures. While the TOJ and Corsi Block cognitive tests had been used previously in such studies (Frenkel & Bourdin, 2009; Lanfranchi, Carretti et al., 2009; Yang, Conners et al., 2014), the difficulty encountered by many children with DS in carrying out these tests led to frustration for both parents and

children. The level of difficulty and the duration of the test were found to be the main challenges.

On reflection, the age profile for both cohorts needs to be reviewed. While the initial recruitment age was 4–8 years for the TD group, the content of the intervention was more appropriate for children aged between 4–6 years. The relevance of this was reflected in the number of TD participants who withdrew from the music programme (6, M=4; F=2), as many of these were aged 7–8. The age range for the DS cohort of 4–12 years was suitable and again reflected in the numbers that engaged with the music programme until the end of the study, with only two withdrawing from the group (M=1; F=1). The dosage length of the intervention of seven weeks was quite limiting. Therefore, a more extended intervention with a shorter weekly engagement dosage might have been optimal. Also, it would have been beneficial to carry out the intervention alongside a contrasting intervention dosage to ascertain the optimum prescribed amount to cause an effect.

Further Studies

Certain aspects of the study design need to be addressed prior to further delivery of the project. The initial two waves of the MINdDS project have acted as a feasibility study, and while it has shown the project's potential, the study design, the content of the intervention and the measures used need to be reconsidered. As recruiting adequate numbers to engage with such studies has proved challenging (Gemma, Pablo et al., 2020), a waitlist control design would be more appropriate with a small sample size rather than having three control conditions. In a waitlist control design, some participants act as a control group and 'wait' to receive the intervention (Patterson et al., 2016). The age profile of the TD cohort needs to be reduced from 4–8 years to 4–6 years, as the material was more appropriate for this age group. The format of the online class would need to be adjusted to increase its accessibility and hence improve children's engagement with this format.

Dosage or the administration of amounts is associated with interventions requiring controlled medication. Until recently, considering dosage in interventions that do not require some medication has received considerably less attention. However, most recently, Frizelle et al. have researched the impact of quantitative dosage on related outcomes in speech and language interventions (Frizelle et al., 2021; Frizelle et al., in print). This study suggests that there is a point where, in this case, vocabulary interventions diminish when there is an additional dosage than what is required (Frizelle et al., 2021). Therefore, this calls into question the optimal dosage levels in intervention studies. Until recently, the relevance of dosage in music has been limited to studies that have been carried out to improve sleep quality (Dickson & Schubert, 2020). Additional studies that have examined the impact of a prescribed amount of music have been in music therapy contexts and how it can increase responses of engagement to treatments in adolescents and adults with mental health concerns (Rodwin et al., 2022). However, 'The feasibility of an online language intervention delivered through music and the impact of dosage on vocabulary outcomes in young children with Down Syndrome' is one of the first studies to explicitly address the impact of music dosage and language development (Frizelle & McMullan et al. in print).

A question of dosage also needs to be considered in relation to future waves of the MINdDS project. For example, it was found that the preferred dosage by parents was 15 minutes four times a week. However, the desired amount of time by parents, while increasing the commitment to the project, may, in fact, not be the optimum amount to get the desired results. Therefore, this intervention model would have benefited from a contrasting dosage to assess the optimum amount of music exposure required to see benefits (Frizelle et al., 2024). As seen in the results, the TD cohort had a contrasting learning trajectory to the MA-matched children with DS. These findings also suggest that perhaps the TD group needed less of an intervention dosage or more varied content to heighten their engagement.

As the instrument parade and song singing with actions were the preferable activities, the content of the intervention could be altered to reflect these preferences while a more engaging way of teaching the rhythm family clocks and the music vocabulary could be created. This could potentially limit attrition in the study.

The selected TOJ and Corsi Blocks test proved to be too long and difficult for the children with DS. Therefore, finding an alternative set of cognitive measures, such as the CANTAB battery of tests (Tristão et al., 2023) or the Arizona Cognitive Test Battery (ACTB) (Edgin et al., 2017), as well as an animated format of the Corsi Blocks, would be more appropriate (Lanfranci, 2014). The CANTAB battery has been used with children with DS and has proven to be an effective measure in assessing key cognitive domains (Tristão et al., 2023). Similarly, the ACTB has also been used with this cohort and has been found effective for testing memory and attention, as well as being suitable for retesting in research studies (Edgin et al., 2017)

In addition, one of the main challenges encountered was the collection of saliva from this cohort, as they have a reduced salivary flow (Areias et al., 2012). Therefore, finding an alternative biological measure to test oxytocin would be preferable. Certain intestinal microbes promote regular gut transit, of which the function of normal social behaviour in mice has been found and impacted by oxytocin in the gut (Danhof et al., 2023). Testing the gut microbiome could be a more accessible sample to obtain from children with DS rather than saliva (Shaoli et al., 2023; Varian et al., 2023). University College Cork is a centre of excellence for research on the gut microbiome (https://www.ucc.ie/en/apc/), and the microbiota

testing of children with DS has not been carried out here thus far. Therefore, this could also be a more accessible way of assessing the effects of music on this cohort that could give a more accurate reading on whether music impacts oxytocin levels and its link to social behaviour and well-being. In addition, part two of the project, which includes EEG assessments, could also be carried out. This would give an additional perspective on the effects of the music intervention on both TD children and those with DS.

Further studies should also include the rollout of the music intervention in school environments, where the learning contexts between the home and school settings could be compared. While the same content would be administered, the learning environments are very different. The school environment would provide a group music learning experience by an education professional. Whereas the learning would be scaffolded individually in the child's home by the parent. This would allow for reviewing the contrasting roles and methodologies undertaken by a parent in the home compared to a school setting and whether one is more effective (Daunhauer, Fidler et al., 2014). This could also contribute to developing more helpful teaching methodologies for this cohort that were acknowledged as lacking by the parents.

As seen in the results section, parents noted marked improvements in communication skills after the music intervention. This has been the focus of an additional study where, in collaboration with Dr. Pauline Frizelle, senior lecturer in Speech and Hearing Sciences at UCC, miniMINdDS was created, a project that examines the impact of a 12-week music intervention programme on language development in children with DS. Twelve videos were created that included singing and signing (Lámh) to over 70 children aged between 1 and 3 years. The results have shown that children who have a critical minimum language level benefit the most from a higher prescribed intervention dosage (Frizelle & McMullan et al., 2024). This research project has received additional funding to progress this study further and address the dire shortage of SLT available to children with DS.

Conclusions

The benefits of music in the developmental trajectory of children have been repeatedly documented and continuously researched (Young, 2023; Selmani, 2024). While there have been substantial studies that focus on the effect of music therapy approaches on children with autism spectrum disorder (ASD) (Ke et al., 2022), the impact of music on the lives of children with DS has received very limited attention (Gemma, Pablo et al., 2020). With an estimated figure of 6 million people with DS worldwide (Hanney, Prasher et al., 2012), there is a distinct void in the reported benefits of music for this population (Gemma, Pablo et al., 2020). Within this study, we aimed to test potential improvements in temporal perception, visual memory and alterations to cortisol and oxytocin after a music intervention with TD children compared to children with DS. However, the biological samples could not be processed, and the results from the cognitive tests showed there was no change post-intervention. Therefore, the conclusions are based almost exclusively on qualitative data.

Recruiting significant numbers from this cohort is particularly challenging, and therefore, finding ways to bring this study to an international level would ensure increased numbers. MINdDS also showed the huge disparity between CA and MA in children with DS and the need to account for this in research projects. Therefore, in agreement with Yang et al., the PPVT is the most effective measure to establish the MA of children with DS, as their receptive language is less impaired (Yang, Conners et al., 2014).

The study has shown that there is a decline in the number of those who avail of music lessons in both TD and DS cohorts. The lack of availability was one of the fundamental reasons why parents and guardians had not enrolled their children in music. However, in addition, the parents of those with DS felt there was an unavailability of suitable music programmes with experienced teachers. These voids helped shape and influence the content and delivery of MINdDS. The content of MINdDS connected musical concepts and ideas to relatable contexts that the children were already familiar with. The children could transfer their music skills to everyday contexts and vice versa. The most significant engagement was found with the

sequential songs and the instrument recognition, which should shape the content of further interventions. In addition, there is a need to consider dosage levels, particularly for the TD cohort, whose engagement climaxed consistently at week six in each component and then declined. This study also solidified the importance of repetition for the DS groups, with marked increases from week to week found across each musical element. The importance of repetition and rehearsal for children with DS is consistent with the literature, particularly in the speech and language development of this cohort (Walsh, 1999; Frizelle, 2017).

The systematic review by Gemma et al. highlighted that existing studies on music and those with DS have concentrated primarily on its impact on social and emotional development (37%) and multiple areas of development (32%) in this cohort (Gemma Pablo et al., 2020). However, areas such as communication development (10%), motor skills (5%), and general cognitive development (16%) have received considerably less attention (Gemma, Pablo et al., 2020). In contrast, the findings in this preliminary study point to the potential significance of music on communication and cognitive skills in this cohort. A more accessible measure of assessing these potential changes in cognition in children with DS would increase compliance and encourage more to be involved in the study. This increase in numbers would enable the processing of biological markers to assess the impact of the music programme on oxytocin and cortisol.

This link between song singing, communication skills, and memory emerged as the main areas that require further investigation. The study also showed the potential of the impact of music on the general quality of life also requires additional research as this study is underpowered. While observations suggest that music could potentially impact emotional functioning, social functioning, and school/work functioning in this cohort, this suggestion remains to be tested.

It was noted in the study that both TD and DS children have very different developmental trajectories. However, developmental models, to date, have not been able to successfully account for atypical developmental trajectories. As has been promoted in this thesis, a child's development should be considered in relation to their own context. Therefore, it is worth considering these developmental changes in behavioural and cognitive outcomes in relation to the neuro-causal model outlined

in the introduction section of this thesis. Proactive engagement, one of the developing processes, as seen in the model, and progress, can influence the cognitive (hot and cold) and behavioural outcomes in a child's development. These outcomes, while impacting how a child progresses, also feed directly back into the contexts of the individual. Consequently, the contexts, specifically the brain, environment, and body, are potentially always in a state of change. Therefore, the derived neuro-causal model that has dominated this thesis is never static, as it promotes a cyclical process of context, developing processes and outcomes constantly changing and impacting development regardless of having an atypical developmental profile. While it has acted as a structuring mechanism throughout this thesis, this model offers further potential that could help view a child's development in relation to their contextual abilities.

Fuzzy Logic: Straddling the Intersection between Music Therapy and Music Education

The preference for music therapy when engaging children with DS in music has become evident throughout this thesis (Gemma, Pablo et al., 2020). Music therapy is of fundamental importance and has been predominantly linked to children with disabilities (Gemma et al., 2020; Mina et al., 2021). It is a welcomed, growing profession and practice that has gained increased acknowledgement (Register, 2013). Recognising this, many parents felt music could support their child's emotional regulation (Moore, 2015). However, there is still much confusion over the role that music therapy has in working with children with disabilities. (Sundar, 2014). In many cases, such as emphasised in this project, an informed music educator equipped with an appropriate skill set can facilitate music learning for all abilities. However, many music practitioners have limited experience teaching music to neurodiverse learners, resulting in frustration for parents, as was found in the contextual content analysis of Why Choose Music? Such practitioners require further support and resources to make their teaching more applicable to these learners (Abad et al., 2023).

Several sources have segregated musical interventions, such as the one proposed by Raglio et al. in 'Music and Health: What Interventions for What Results?'. Raglio et al. appropriately distinguish between interventions by trained credentialed

music therapists, listening-to-music, and music-based approaches (Raglio et al., 2011). However, MINdDS has merit outside of a purely therapeutic perspective and more scientific research than the general music-based approaches that Raglio suggests (Raglio et al.,2011). For example, under the general music-based approaches, the absence of an intervention model and the eradication of psychological and neuroscientific models are assumed (Raglio et al., 2011). As seen throughout this thesis, the MINdDS project has been framed by a neuro-causal model (Cebula, 2010; Westermann, 2010). Moreover, it has a detailed pedagogical practice informed by neuroscience, music education, and psychology. Qualified music practitioners and educators have delivered the music intervention. The YouTube videos and online classes embrace therapeutic elements; however, the content is framed by music education.

Therefore, adopting a binary view of music therapy versus other music interventions or practices only compounds the fuzzy logic between music therapy and educative practices. There is a dire need for a redefined approach to music interventions outside the realm of music therapy, giving the professionals working in that area recognition for the valued work underscored by detailed research and expertise. This area of growth needs to be further explored in an interdisciplinary field of music interventions that, while containing therapeutic elements, does not strive to implement the additional responsibilities of the music therapy but still has merit in the work being done. Perhaps adopting a model of detailed analysis of these music progammes to 'Protect the Power of Interventions through proper reporting' (Conn & Groves, 2011). This could be a way to move the research forward (Conn & Groves, 2011). Involving interdisciplinary partners in this process would inform the creation of more appropriate and effective music interventions for our wonderfully talented and creative children with DS so that we can empower these children to celebrate their immense abilities (Zhang et al., 2023).

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Appendix A: Information Leaflets, Consent, and Assent Forms



INFORMATION LEAFLET

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

FOR THE PARENTS OF CHILDREN WITH DOWN SYNDROME ENGAGING WITH MUSIC TRAINING

THE MUSIC4CHILDREN PROGRAMME

Music4Children is a music programme that was devised in 2011 to introduce children to music through fun and game activities. Inspired by her own children, Dr. Eva McMullan wanted to emphasise the importance of music in the development of children and their overall well-being. Since their musical beginnings, Eva, in association with her colleague Padraig Wallace, has developed a programme that celebrates music learning by taking inspiration from the children they work with. An integral element of the programme is accessibility and see children of differing abilities learn together in an inclusive environment. *"I don't think any of us as parents at Down Syndrome Cork, when we started music with Eva and the puppets at Music4Children ever imagined the impact it would have on our kids. The amount of benefits these sessions have on auditory processing, language skills, speech, shared experience with parents, attention building, turn taking, literacy skills and most of all fun is astounding."* (Alison Wilson- Conor's Mum)

THE RESEARCH

The impact of this programme has been acknowledged by both parents and children and, therefore, has been integral in encouraging further research into the benefits of music education on child development. This impact has been particularly prevalent in children with Down Syndrome. In association with the School of Applied Psychology, Department of Anatomy and Neuroscience and the Department of Music UCC, Down Syndrome Cork and Music4Children, the benefits of music engagement on the neurologic development of children with Down Syndrome will be investigated. This is important because, while the reports of the parents involved are very

important, we need to see if such benefits can be detected with clinical tests, and this intervention can be useful for children with different difficulties.

WHAT IS INVOLVED?

This study will require you and your child to attend an online pre-training testing for one hour as well as respond to some questions on music engagement in a survey. Then, you will be required to engage with the music video on a daily basis in your home and be part of an online music class every Saturday for a 45-minute class for eight weeks. You will be sent a music bag with the relevant materials in advance of the study.

During this time, the child will engage with the fundamentals of music, including sequential songs, rhythm and melody games and learn about different instruments. This will involve the use of voice, movement, and simple musical tools. The post-training testing will involve a second 1-hour online session. The testing process is detailed in the material that follows.

TESTING

The children will be mentally-aged matched to a control group of typically developing children by using the The Peabody Picture Vocabulary Test. This is an assessment tool for mental age widely used by clinicians and researchers; the test requires to match words to pictures and cover our selected age range. It takes approximately half an hour and will take place online. The additional testing will include collection of saliva and hair for measurement of hormone levels, online computergames and memory-based tasks. Hormone levels can assist in assessing the benefits of musical engagement. The participants and their parents will be told how to collect saliva at an information evening that will be held on Microsoft Teams in advance of the study. 8 salivettes will be given to each participant, 4 to be used at the start of the study and 4 to be used at the end of the study. Samples will be collected: immediately on waking, 15, 30 and 45 minutes after waking. Parents will also collect hair samples before and after the study. This will be done by using a scissors as close to the scalp as possible and will involve 50-100 strands of hair. Both hair and saliva samples will be collected by the researcher from the participants' homes. Additional testing will include some online computer-based and game-like cognitive tests that examine the impact of music on memory.

QUESTIONNAIRES

You, the parent, will be required to fill out two questionnaires online. 'Why Choose Music?' will be filled out at the commencement of the study. The PedsQL Quality of Life Inventory will be filled before the study and when the music training has been completed. A short daily diary will also be required to be kept during the 8 weeks of music training.

BENEFITS

It is important to emphasise that while children benefited from Music4Children, we cannot promise that your child will also benefit, we will not be sure until we try. In any case, you will contribute to understanding whether this programme works and how we can improve it. While the importance of music as an effective therapeutic tool has received support both within applied musicological and medical contexts,

the awareness of music training and education as having an impact on the neurocognitive development of children and adults with intellectual impairments remains under-explored. Understanding the impact of music engagement is critically important to understand how it can be used in conjunction with therapeutic interventions. Additionally, this will allow us to refine the programme to either modulate neural activity by adjusting the behavioural training.





FOR FURTHER INFORMATION

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Tel: 021 494530

Celebrating Music Learning through fun and games by taking inspiration from children themselves.



INFORMATION LEAFLET

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

FOR THE PARENTS OF CHILDREN WITH DOWN SYNDROME NOT ENGAGING WITH MUSIC TRAINING

THE MUSIC4CHILDREN PROGRAMME

Music4Children is a music programme that was devised in 2011 to introduce children to music through fun and game activities. Inspired by her own children, Dr. Eva McMullan wanted to emphasise the importance of music in the development of children and their overall well-being. Since their musical beginnings, Eva, in association with her colleague Padraig Wallace, has developed a programme that celebrates music learning by taking inspiration from the children they work with. An integral element of the programme is accessibility and see children of differing abilities learn together in an inclusive environment. *"I don't think any of us as parents at Down Syndrome Cork, when we started music with Eva and the puppets at Music4Children ever imagined the impact it would have on our kids. The amount of benefits these sessions have on auditory processing, language skills, speech, shared experience with parents, attention building, turn taking, literacy skills and most of all fun is astounding."* (Alison Wilson- Conor's Mum)

THE RESEARCH

The impact of this programme has been acknowledged by both parents and children and, therefore, has been integral in encouraging further research into the benefits of music education on child development. This impact has been particularly prevalent in children with Down Syndrome. In association with the School of Applied Psychology, Department of Anatomy and Neuroscience and the Department of Music UCC, Down Syndrome Cork and Music4Children, the benefits of music engagement on the neurologic development of children with Down Syndrome will be investigated. This is important because, while the reports of the parents involved are very important, we need to see if such benefits can be detected with clinical tests, and this intervention can be useful for children with different difficulties.

WHAT IS INVOLVED?

In order to see if there are changes in the brains of children getting musical training, we need to compare them to children that are not receiving any musical training.
Your child has been placed in the group that will not receive musical training. This is because they are not currently enrolled in any music classes. This study will require you and your child to attend an online pre-training testing for one hour, as well as respond to some questions on music engagement in a survey, and again after an eight-week period. The testing process is detailed in the material that follows. There is nothing for you to do between these two visits.

TESTING

The children will be mentally-aged matched to a control group of typically developing children by using the The Peabody Picture Vocabulary Test. This is an assessment tool for mental age widely used by clinicians and researchers; the test requires to match words to pictures and cover our selected age range. It takes approximately half an hour and will be carried out online. The additional testing will include collection of saliva and hair for measurement of hormone levels, online computer games and memory-based tasks. Hormone levels can assist in assessing the benefits of musical engagement. The participants and their parents will be told how to collect saliva at an information evening that will be held on Microsoft Teams in advance of the study. 8 salivettes will be given to each participant, 4 to be used at the start of the study and 4 to be used at the end of the study. Samples will be collected: immediately on waking, 15, 30 and 45 minutes after waking. Parents will also collect hair samples before and after the study. This will be done by using a scissors as close to the scalp as possible and will involve 50-100 strands of hair. Both hair and saliva samples will be collected by the researcher from the participants' home. Additional testing will include some online computer-based and game-like cognitive tests that examine the impact of music on memory.

QUESTIONNAIRES

You, the parent, will be required to fill out two questionnaires online. 'Why Choose Music?' will be filled out at the commencement of the study. The PedsQL Quality of Life Inventory will be filled out also at the start of the study.

BENEFITS

While the importance of music as an effective therapeutic tool has received support both within applied musicological and medical contexts, the awareness of music training and education as having an impact on the neurocognitive development of children and adults with intellectual impairments remains under-explored. Understanding the impact of music engagement is critically important to understand how it can be used in conjunction with therapeutic interventions. Additionally, this will allow us to refine the programme to either modulate neural activity by adjusting the behavioural training.





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INFORMATION LEAFLET

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

FOR THE PARENTS OF TYPICALLY DEVELOPING CHILDREN ENGAGING WITH MUSIC TRAINING

THE MUSIC4CHILDREN PROGRAMME

Music4Children is a music programme that was devised in 2011 to introduce children to music through fun and game activities. Inspired by her own children, Dr. Eva McMullan wanted to emphasise the importance of music in the development of children and their overall well-being. Since their musical beginnings, Eva, in association with her colleague Padraig Wallace, has developed a programme that celebrates music learning by taking inspiration from the children they work with. An integral element of the programme is accessibility and see children of differing abilities learn together in an inclusive environment. *"I don't think any of us as parents at Down Syndrome Cork, when we started music with Eva and the puppets at Music4Children ever imagined the impact it would have on our kids. The amount of benefits these sessions have on auditory processing, language skills, speech, shared experience with parents, attention building, turn taking, literacy skills and most of all fun is astounding."* (Alison Wilson- Conor's Mum)

THE RESEARCH

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we need to see if such benefits can be detected with clinical tests and this intervention can be useful for children with different difficulties.

WHAT IS INVOLVED?

This study will require you and your child to attend an online pre-training testing for one hour as well as respond to some questions on music engagement in a survey. Then, you will be required to engage with the music video on a daily basis in your home and be part of an online music class every Saturday for a 45-minute class for eight weeks. You will be sent a music bag with the relevant materials in advance of the study.

During this time the child will engage with the fundamentals of music, including sequential songs, rhythm and melody games and learn about different instruments. This will involve the use of voice, movement and simple musical tools. The post-training testing will involve a second 1-hour online session. The testing process is detailed in the material that follows.

TESTING

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School of
Applied Psychology

Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

PARENT INFORMATION/CONSENT FORM FOR CHILDREN WITH DOWN SYNDROME ENGAGING WITH MUSIC

CONSENT BY SUBJECT FOR PARTICIPATION IN RESEARCH PROTOCOL

Section A

Protocol Number:_____ Participant Name:_____

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

Investigators Directing Research:
Chief Investigator: Dr. Jason S. Chan,
Researchers: Dr Annalisa Setti,
Dr. Yvonne Nolan,
Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904530

Your child is being asked to participate in a research study that will assess how daily interaction with a specifically designed music programme will impact the neurocognitive development of children with Down Syndrome and typically developing children. In order to decide whether or not you and your child want to be a part of this research study, you should understand enough about its risks and benefits to make an informed judgment. This process is known as informed consent. This information and consent form gives detailed information about the research

study, which will be discussed with you. Once you understand the study, you will be asked to sign this form if you wish your child to participate.

Section B

I. WHAT YOU AND YOUR CHILD WILL BE ASKED TO DO?

The aims of this project are to understand the behavioural, neural, and hormonal correlates associated with musical training and to establish whether it can be used as a possible intervention for children with Down Syndrome. While Down Syndrome is an incurable genetic disorder, early behavioural intervention can significantly improve independence in later life, we aim to understand whether Musical training constitute a potential way to obtain improvement in the short term, i.e. by the end of the training.

If you agree for your child to participate in the study, he/she will be required to engage with a music programme for eight weeks. This will include watching a video at home on daily basis and engaging with a weekly 45-minute music class online. This music programme will introduce the children to the fundamentals of music and will include sequential songs, rhythm and melody games and learning about musical instruments. This will involve the use of voice, movement, and simple musical tools. You will be sent a music bag in advance of the study.

You and Your child will also be required to engage with various tests, in two separate online testing sessions. The initial test will allow the investigators to mentally age match the children by using the Peabody Picture Vocabulary Test. This is an assessment tool for mental age widely used by clinicians and researchers; the test requires to match words to pictures and cover our selected age range. It takes approximately half an hour online.

The additional testing will include playing online computer games designed to evaluate cognition. Saliva testing will also be carried out and will include collecting saliva samples at home. In addition, hair samples will be collected at the first and final study visit. Both of these samples will be collected by the researcher at the start of the study and at the conclusion of the study to help assess the effectiveness of the music intervention. An information evening will be held on Microsoft Teams in advance of the study to show you how to collect the hair and saliva samples.

Parents will collect the saliva samples from their child at home. To do this you will be given 8 salivettes, 4 to be used at the start of the study and 4 to be used at the end of the study. These salivettes are plastic containers containing a swab. These have been age-appropriately designed for ease of collection from children. Samples will be collected immediately on waking, and then again at 15, 30 and 45 minutes after waking on one morning just before the start of the study and again one day at the end of the study. Levels of the hormones cortisol and oxytocin will be analysed in the laboratory from the saliva samples. The saliva sample must be stored at 4°C until

collected by the researcher. Because cortisol accumulates in hair over time, we will also collect hair samples at the beginning and at the end of the study (a very small sample). A hair sample will be collected by you the parent using scissors as close to the scalp as possible at the first and last study visit. The sample will be wrapped in tin foil and stored at -20°C until collected by the researcher and cortisol concentration is measured in the lab.

You will be required to fill out two online questionnaires before the commencement of the study to establish basic details prior to the study. One questionnaire is entitled 'Why Choose Music?' that helps to establish details of your child's prior engagement with music, if any and basic demographic information. The second questionnaire is the 'Paediatric Quality of Life Inventory' in which you are invited to answer questions to measure health-related quality of life. This will be filled out at the start and the concluding point of the study to check for any change after the 8 weeks. In addition, you will be required to keep a reflective diary throughout the 8 weeks of musical training.

II. WHAT ARE THE POTENTIAL RISKS AND BENEFITS?

There is no risk of harm to children: the equipment used, and the tests are non-harmful and have been traditionally used with various healthy and pathological groups in the literature; precaution will be taken to ensure that children are not excessively fatigued and distressed because of the length of the tests. All precautions have been taken to minimise risk. There will be the opportunity for breaks about every 5-7 minutes during the tests, but the testing can be paused at any point to ensure the comfort and well-being of the contributor. In terms of musical training, there is no risk of harm, the training has been delivered to children with and without disability in Cork and implies the use of voice, movement, and simple harmless musical tools.

The main benefit of this research will hopefully allow us to refine music training programmes that will impact positively on children with Down Syndrome or those that have other forms of cognitive impairments.

III. FURTHER DETAILS

Participation is voluntary. If you and your child decide to take part in the study and then change your mind, you and your child can withdraw from the study at any point, and all their data will be permanently deleted. Behavioural, EEG and saliva/hair testing are independent components of the study; therefore, should one of them be distressing for a child, it can be dismissed. You and your child may also decide not to take part in this study; this will not affect your ability to take part in the classes.

The tests will be carried out by Dr. Eva McMullan, Dr. Jason S. Chan, Dr. Annalisa Setti, and Dr. Yvonne Nolan. Eva has completed a first aid course and a child protection course, and all investigators have obtained Garda Clearance. All information collected

will not be associated with your child's name. When the results are published, it will be impossible to identify individual participants. All data will be securely stored on a password-protected computer, and any information about participant's identities will be removed. The results will be presented in an MSc thesis prepared by Dr. McMullan. They will be seen by the supervisors (named above), an internal examiner and an external examiner in accordance with university regulations. The study may be read by future students and may be published in an academic journal. The data collected will remain on record for a period of at least 10 years. Only the investigators in this research project will have access to this material during this time. They are Dr. Jason S. Chan, Dr. Yvonne Nolan, Dr. Annalisa Setti and Dr. Eva McMullan-Glossop.

Section C AGREEMENT TO CONSENT

The research project and the treatment procedures associated with it have been fully explained to me. All experimental procedures have been identified and no guarantee has been given about the possible results. I have had the opportunity to ask questions concerning any and all aspects of the project and any procedures involved. I am aware that participation is voluntary and that I may withdraw my consent for my child to participate at any time. I am aware that my decision not to participate or to withdraw will not restrict my child's access to music training normally available to me. Confidentiality of records concerning my involvement in this project will be maintained in an appropriate manner.

I understand that the investigators have such insurance as is required by law in the event of injury resulting from this research.

I, the undersigned, hereby consent my child to participate as a subject in the above-described project conducted at the School of Applied Psychology, Department of Music, and Department of Anatomy and Neuroscience.

I have received a copy of this consent form for my records. I understand that if I have any questions concerning this research, I can contact the researchers listed above. If I have further queries concerning my rights in connection with the research, I can contact the Clinical Research Ethics Committee of the Cork Teaching Hospitals, Lancaster Hall, 6 Little Hanover Street, Cork.

After reading the entire consent form, if you have no further questions about giving consent, please sign where indicated.

Investigator:

Name of Child

Name of Parent/Guardian:

O I consent for my child to begin the study

O I do not consent; I do not wish my child to participate in the study

Please select the date

	Month	Day	Year
Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)





UCC
University College Cork, Ireland
Coláiste na hOllscoile Corcaigh

School of
Applied Psychology

Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

DATA PROTECTION NOTICE

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

At University College Cork, we treat your privacy seriously. Any personal data which you provide to the University will be treated with the highest standards of security and confidentiality in accordance with Irish and European Data Protection legislation. This notice sets out details of the information that we collect, how we process it and who we share it with. It also explains your rights under data protection law in relation to our processing of your data.

Who we are

Throughout this Notice, "we", "us", and "our" refer to University College Cork as the study sponsor. For more information about us, please refer to our website: www.ucc.ie

How we will use your personal data

By participating in the study, information about your child (also called "personal data") will be collected for the study purposes mentioned in the Patient information leaflet. This personal data includes, for example:

- information that directly identifies your child (such as their name and year of birth).
- their gender, ethnic and racial background.
- information on their health, medical condition, and quality of life.
- information on their previous music education.

- information on their cognitive pre-tests and post-tests, including the Peabody Vocabulary Test.
- information contained in their hair and saliva samples and the results after analysis.

Personal data collected at any time during the study will be kept strictly confidential. To ensure confidentiality, the data generated during the study is **coded** with a number that will identify you in the study. Any information that leaves the clinical site will be labelled with your code instead of your name. Every person who has access to your uncoded data (that is kept at the School of Applied Psychology, University College Cork) is subject to professional secrecy and confidentiality.

Data that directly identifies you (uncoded data) is stored in your medical files at the clinical site in which the data have been obtained. The 'key' that connects the subject name and subject ID should be kept in a separate location to the data; securely (locked cupboard in a room with restricted access) by the investigator. All files which contain personally identifiable information (e.g., keys) should undergo an additional layer of encryption such as Microsoft

Azure Information Protection.
Unencrypted personal devices (laptops or phones) should not be used to acquire data.

Who will access my personal data?

Your uncoded data will only be accessible to the study investigator and staff. The study's results will be provided to the Clinical Research Ethics Committee of the Cork Teaching Hospitals (CREC) in compliance with national and international regulations on clinical studies.

The purpose and legal basis for collecting your data.

Any personal data you provide to us during the course of this study will be processed fairly and lawfully.

Signing the Informed Consent Form means that your personal data will be used for the purposes outlined in the Patient information leaflet (PIL).

Personal data collected during this study and the results of the study may be presented for scientific purposes. However, you will never be identified individually during these presentations. Your identity will not be revealed in any reports or publications.

The clinical site and the investigator will use your personal data within the scope defined above.

The General Data Protection Regulation allows us to process your data because you have provided your consent. You are entitled to withdraw your consent at any time.

How long we will keep your data.

The personal data collected in the study will be kept for a period of 10 years after the end of the study. However, your anonymised data will be held in accordance with FAIR data principles and may be stored indefinitely in an open data repository.

Your rights

You have various rights under data protection law, subject to certain exemptions, in connection with our processing of your personal data, including the right:

- to find out if we use your personal data, access your personal data, and receive copies of your personal data.
- to have inaccurate/incomplete information corrected and updated.
- in certain circumstances, to have your details deleted from systems that we use to process your personal data or have the use of your personal data restricted in certain ways.
- to object to certain processing of your data by UCC.
- to exercise your right to data portability where applicable (i.e. obtain a copy of your personal data in a commonly used electronic form.
- to withdraw your consent to the processing of your data up to 30 days of participation without giving a reason by notifying your decision to the investigator. This will not affect the lawfulness of processing data about you based on your consent before the withdrawal. If you withdraw your consent for data processing, your participation in the study stops and no further data will be collected from you. Your study physician will present you the options you have concerning your personal data.
- along with study withdrawal, you have the right to request the deletion of data about you, up to 30 days of participation, if your data are no longer necessary for the purposes of processing or there is no other legal ground for their further processing.
- after 30 days of participation, the link between your personal identity and your data will be destroyed, and we will no longer be able to identify you from the rest of the dataset. Thus, it will not be possible to withdraw your data.

If you wish to exercise any of these rights, please address your request to the study coordinator or the Information Compliance Manager University College Cork (details below).

Questions or Complaints

If you have any queries in relation to this study, please contact Dr. Eva McMullan at e.mcmullan@ucc.ie.

If you have any complaints in connection with our processing of your personal data, you can contact UCC's Information Compliance Manager: **Information Compliance Manager, Office of Corporate & Legal Affairs, University College Cork, Western Road, Cork E: foi@ucc.ie Tel: +353 21 4903949**

You also have the right to lodge a complaint with the Data Protection Commission if you are unhappy with our processing of your personal data. Details of how to lodge a complaint can be found on the Data Protection Commission's website www.dataprotection.ie) or by telephoning 1890 252 231.

Name of Parent/Guardian _____

Date _____

Name of Informant _____

Date _____

In submitting this form, you understand and agree that your personal details will be used by the School of Applied Psychology for the purposes outlined above.

Submit



UCC
University College Cork, Ireland
Coláiste na hOllscoile Corcaigh

School of
Applied Psychology

Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

PARENT INFORMATION/CONSENT FORM FOR CHILDREN WITH DOWN SYNDROME NOT ENGAGING WITH MUSIC

CONSENT BY SUBJECT FOR PARTICIPATION IN RESEARCH PROTOCOL

Section A

Protocol Number:_____ Participant Name:_____

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

Investigators Directing Research:
Chief Investigator: Dr. Jason S. Chan,
Researchers: Dr Annalisa Setti,
Dr. Yvonne Nolan,
Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904530

Your child is being asked to participate in a research study that will assess how daily interaction with a specifically designed music programme will impact on the neurocognitive development of children with Down Syndrome and typically developing children. In order to decide whether or not you want to be a part of this research study, you should understand enough about its risks and benefits to make an informed judgment. This process is known as informed consent. This information

and consent form gives detailed information about the research study, which will be discussed with you. Once you understand the study, you will be asked to sign this form if you wish your child to participate.

Section B

I. WHAT YOU AND YOUR CHILD WILL BE ASKED TO DO?

If you agree that your child will participate in the study, he/she will be required to engage with various tests in two separate online testing sessions. The initial test will allow the investigators to mentally age-match the children using the Peabody Picture Vocabulary Test. This is an assessment tool for mental age widely used by clinicians and researchers; the test requires to match words to pictures and cover our selected age range. It takes approximately half an hour online. The additional testing will include computer games.

Saliva testing will also be carried out and will include collecting saliva samples at home. In addition, hair samples will be collected before the study begins. Both of these samples will be collected by the researcher at the start of the study and at the conclusion of the study. All tests will be carried at the commencement of the study and once again after an eight-week period. An information evening will be held on Microsoft Teams in advance of the study to show you how to collect the hair and saliva samples. There is nothing your child needs to do between these testing sessions.

Parents will collect the saliva samples from their child at home. To do this, you will be given 8 salivettes, 4 to be used at the start of the study and 4 to be used at the end of the study. These salivettes are plastic containers containing a swab. These have been age-appropriately designed for ease of collection from children. Samples will be collected immediately on waking and then again at 15, 30 and 45 minutes after waking on one morning just before the start of the study and again one day at the end of the study. Levels of the hormones cortisol and oxytocin will be analysed in the laboratory from the saliva samples. The saliva sample must be stored at 4°C until collected by the researcher. Because cortisol accumulates in hair over time, we will also collect hair samples at the beginning and at the end of the study (a very small sample). A hair sample will be collected by you the parent using scissors as close to the scalp as possible at the first and last study visit. The sample will be wrapped in tin foil and stored at -20°C until collected by the researcher and cortisol concentration is measured in the lab.

You will be required to fill out two online questionnaires before the commencement of the study to establish basic details prior to the study. One questionnaire is entitled ‘Why Choose Music?’ that helps to establish details of your child’s prior engagement with music, if any and basic demographic information. In the second questionnaire, the ‘Paediatric Quality of Life Inventory, you are invited to answer questions to

measure health-related quality of life prior to the study. You will be again invited to answer the 'Paediatric Quality of Life Inventory' after an eight-week period.

II. WHAT ARE THE POTENTIAL RISKS AND BENEFITS?

There is no risk of harm for children: the equipment used and the tests are non-harmful and have been traditionally used with various healthy and pathological groups in the literature; precaution will be taken to ensure that children are not excessively fatigued and distressed as a consequence of the length of the tests. All precautions have been taken to minimise risk. There will be the opportunity for breaks about every 5-7 minutes during the tests, but the testing can be paused at any point to ensure the comfort and well-being of the contributor.
The main benefit of this research will hopefully allow us to refine music training programmes that will positively impact children with Down Syndrome or those with other forms of cognitive impairment.

III. FURTHER DETAILS

Participation is voluntary. If you and your child decide to take part in the study and then change your mind, you and your child can withdraw from the study at any point, and all their data will be permanently deleted. Behavioural, EEG and saliva/hair testing are independent components of the study; therefore, should one of them be distressing for a child, it can be dismissed. You and your child may also decide not to take part in this study, this will not affect your ability to take part in the classes.

The tests will be carried out by Dr. Eva McMullan, Dr. Jason S. Chan, Dr. Annalisa Setti, and Dr. Yvonne Nolan. Eva has completed a first aid course and a child protection course, and all investigators have obtained Garda Clearance. All information collected will not be associated with your child's name. When the results are published, it will be impossible to identify individual participants. All data will be securely stored on a password-protected computer, and any information about participant's identities will be removed. The results will be presented in an MSc thesis prepared by Dr. Eva McMullan. They will be seen by the supervisors (named above), an internal examiner and an external examiner in accordance with University Regulations. The study may be read by future students studying this course and may be published in an academic journal. The data collected will remain on record for a period of at least 10 years. Only the investigators in this research project will have access to this material during this time. They are Dr. Jason S. Chan, Dr. Yvonne Nolan, Dr. Annalisa Setti, and Dr. Eva McMullan-Glossop.

Section C AGREEMENT TO CONSENT

The research project and the treatment procedures associated with it have been fully explained to me. All experimental procedures have been identified and no guarantee has been given about the possible results. I have had the opportunity to ask questions

concerning any and all aspects of the project and any procedures involved. I am aware that participation is voluntary and that I may withdraw my consent for my child to participate at any time. I am aware that my decision not to participate or to withdraw will not restrict my child's access to musical training normally available to me. The confidentiality of my records concerning my involvement in this project will be maintained in an appropriate manner.

I understand that the investigators have insurance as required by law in the event of injury resulting from this research.

I, the undersigned, hereby consent to my child's participation as a subject in the above-described project conducted at the School of Applied Psychology, Department of Music, and Department of Anatomy and Neuroscience.

I have received a copy of this consent form for my records. I understand that if I have any questions concerning this research, I can contact the researchers listed above. If I have further queries concerning my rights in connection with the research, I can contact the Clinical Research Ethics Committee of the Cork Teaching Hospitals, Lancaster Hall, 6 Little Hanover Street, Cork.

After reading the entire consent form, if you have no further questions about giving consent, please sign where indicated.

Investigator:

Name of Child:

Signature of Parent/Guardian:

O I consent for my child to begin the study

O I do not consent; I do not wish my child to participate in the study

Please select the date

	Month	Day	Year
Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)



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School of
Applied Psychology

Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

DATA PROTECTION NOTICE

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

At University College Cork, we treat your privacy seriously. Any personal data which you provide to the University will be treated with the highest standards of security and confidentiality in accordance with Irish and European Data Protection legislation. This notice sets out details of the information that we collect, how we process it and who we share it with. It also explains your rights under data protection law in relation to our processing of your data.

Who we are.

Throughout this Notice, "we", "us" and "our" refers to University College Cork, as study sponsor. For more information about us, please refer to our website: www.ucc.ie

How we will use your personal data

By participating in the study, information about your child (also called "personal data") will be collected for the study purposes mentioned in the Patient information leaflet. This personal data includes, for example:

- information that directly identifies your child (such as their name and their year of birth).
- their gender, ethnic and racial background.
- information on their health, medical condition, and quality of life.
- information on their previous music education.

- information on their cognitive pre-tests and post-tests, including the Peabody Vocabulary Test.
- information contained in their hair and saliva samples and the results after analysis.

Personal data collected at any time during the study will be kept strictly confidential. To ensure confidentiality, the data generated during the study is **coded** with a number that will identify you in the study. Any information that leaves the clinical site will be labelled with your code instead of your name. Every person that has access to your uncoded data (that is kept at the School of Applied Psychology, University College Cork) is subject to professional secrecy and confidentiality.

Data that directly identifies you (uncoded data) is stored in your medical files at the clinical site in which the data have been obtained. The 'key' that connects the subject name and subject ID should be kept in a separate location to the data; securely (locked cupboard in a room with restricted access) by the investigator. All files which contain personally identifiable information (e.g., keys) should undergo an additional layer of encryption such as Microsoft Azure Information Protection

Unencrypted personal devices (laptop or phone) should not be used to acquire data.

Who will access my personal data?

Your uncoded data will only be accessible to the study investigator and study staff. Results of the study will be provided to the Clinical Research Ethics Committee of the Cork Teaching Hospitals (CREC) in compliance with national and international regulations on clinical studies.

The purpose and legal basis for collecting your data.

Any personal data you provide to us during the course of this study will be processed fairly and lawfully.

Signing the Informed Consent Form means that your personal data will be used for the purposes outlined in the Patient information leaflet (PIL).

Personal data collected during this study and the results of the study may be presented for scientific purposes. However, you will never be identified individually during these presentations. Your identity will not be revealed in any reports or publications.

The clinical site and the investigator will use your personal data within the scope defined above.

The General Data Protection Regulation allows us to process your data because you have provided your consent. You are entitled to withdraw your consent at any time.

How long we will keep your data.

The personal data collected in the study will be kept for 10 years after the end of the study. However, your anonymised data will be held in accordance with FAIR data principles and may be stored indefinitely in an open data repository.

Your rights

You have various rights under data protection law, subject to certain exemptions, in connection with our processing of your personal data, including the right:

- to find out if we use your personal data, access your personal data, and receive copies of it.
- to have inaccurate/incomplete information corrected and updated.
- in certain circumstances, to have your details deleted from systems that we use to process your personal data or have the use of your personal data restricted in certain ways.
- to object to certain processing of your data by UCC.
- to exercise your right to data portability where applicable (i.e. obtain a copy of your personal data in a commonly used electronic form.
- to withdraw your consent to the processing of your data up to 30 days of participation without giving a reason by notifying your decision to the investigator. This will not affect the lawfulness of processing data about you based on your consent before the withdrawal. If you withdraw your consent for data processing, your participation in the study stops and no further data will be collected from you. Your study physician will present you the options you have concerning your personal data.
- along with study withdrawal, you have the right to request the deletion of data about you, up to 30 days of participation, if your data are no longer necessary for the purposes of processing or there is no other legal ground for their further processing.
- after 30 days of participation, the link between your personal identity and your data will be destroyed, and we will no longer be able to identify you from the rest of the dataset. Thus, it will not be possible to withdraw your data.

If you wish to exercise any of these rights, please address your request to the study coordinator or the Information Compliance Manager University College Cork (details below).

Questions or Complaints

If you have any queries in relation to this study, please contact Dr. Eva McMullan e.mcmullan@ucc.ie.

If you have any complaints in connection with our processing of your personal data, you can contact UCC's Information Compliance Manager: **Information Compliance Manager, Office of Corporate & Legal Affairs, University College Cork, Western Road, Cork E: foi@ucc.ie Tel: +353 21 4903949**

If you are unhappy with our processing of your personal data, you have the right to lodge a complaint with the Data Protection Commission. Details of how to lodge a complaint can be found on the Data Protection Commission's website (HYPERLINK "http://www.dataprotection.ie") or by telephoning 1890 252 231.

Name of Parent/Guardian _____

Date _____

Name of Informant _____

Date _____

In submitting this form, you understand and agree that your personal details will be used by the School of Applied Psychology for the purposes outlined above.

Submit



PARENT INFORMATION/CONSENT FORM FOR TYPICALLY DEVELOPING CHILDREN ENGAGING WITH MUSIC

CONSENT BY SUBJECT FOR PARTICIPATION IN RESEARCH PROTOCOL

Section A

Protocol Number:_____ Participant Name:_____

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

Investigators Directing Research:

Chief Investigator: Dr. Jason S. Chan,
Researchers: Dr Annalisa Setti,
Dr. Yvonne Nolan,
Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904530

Your child is being asked to participate in a research study that will assess how daily interaction with a specifically designed music programme will impact the neurocognitive development of children with Down Syndrome and typically developing children. In order to decide whether or not you and your child want to be a part of this research study, you should understand enough about its risks and benefits to make an informed judgment. This process is known as informed consent. This information and consent form gives detailed information about the research

study, which will be discussed with you. Once you understand the study, you will be asked to sign this form if you wish your child to participate.

Section B

I. WHAT YOU AND YOUR CHILD WILL BE ASKED TO DO?

The aims of this project are to understand the behavioural, neural, and hormonal correlates associated with musical training and to establish whether it can be used as a possible intervention for children with Down Syndrome. While Down Syndrome is an incurable genetic disorder, early behavioural intervention can significantly improve independence in later life. We aim to understand whether Musical training constitutes a potential way to obtain improvement in the short term, i.e. by the end of the training.

If you agree for your child to participate in the study, he/she will be required to engage with a music programme for eight weeks. This will include watching a video at home on daily basis and engaging with a weekly 45-minute music class online. This music programme will introduce the children to the fundamentals of music and will include sequential songs, rhythm and melody games and learning about musical instruments. This will involve the use of voice, movement, and simple musical tools. You will be sent a music bag in advance of the study.

You and your child will also be required to engage with various tests, in two separate online testing sessions. The initial test will allow the investigators to mentally age match the children by using the Peabody Picture Vocabulary Test. This is an assessment tool for mental age widely used by clinicians and researchers; the test requires to match words to pictures and cover our selected age range. It takes approximately half an hour online.

The additional testing will include playing online computer games designed to evaluate cognition. Saliva testing will also be carried out and will include collecting saliva samples at home. In addition, hair samples will be collected at the first and final study visit. Both of these samples will be collected by the researcher at the start of the study and at the conclusion of the study to help assess the effectiveness of the music intervention. An information evening will be held on Microsoft Teams in advance of the study to show you how to collect the hair and saliva samples.

Parents will collect the saliva samples from their child at home. To do this you will be given 8 salivettes, 4 to be used at the start of the study and 4 to be used at the end of the study. These salivettes are plastic containers containing a swab. These have been age-appropriately designed for ease of collection from children. Samples will be collected immediately on waking, and then again at 15, 30 and 45 minutes after waking on one morning just before the start of the study and again one day at the end of the study. Levels of the hormones cortisol and oxytocin will be analysed in the laboratory from the saliva samples. The saliva sample must be stored at 4°C until collected by the researcher. Because cortisol accumulates in hair over time, we will

also collect hair samples at the beginning and at the end of the study (a very small sample). A hair sample will be collected by you the parent using scissors as close to the scalp as possible at the first and last study visit. The sample will be wrapped in tin foil and stored at -20°C until collected by the researcher and cortisol concentration is measured in the lab.

You will be required to fill out two online questionnaires before the commencement of the study to establish basic details prior to the study. One questionnaire is entitled 'Why Choose Music?' that helps to establish details of your child's prior engagement with music, if any and basic demographic information. The second questionnaire is the 'Paediatric Quality of Life Inventory' in which you are invited to answer questions to measure health-related quality of life. This will be filled out at the start and the concluding point of the study to check for any change after the 8 weeks. In addition, you will be required to keep a reflective diary throughout the 8 weeks of musical training.

II. WHAT ARE THE POTENTIAL RISKS AND BENEFITS?

There is no risk of harm for children: the equipment used and the tests are non-harmful and have been traditionally used with various healthy and pathological groups in the literature; precaution will be taken to ensure that children are not excessively fatigued and distressed because of the length of the tests. All precautions have been taken to minimise risk. There will be the opportunity for breaks about every 5-7 minutes during the tests, but the testing can be paused at any point to ensure the comfort and well-being of the contributor. In terms of musical training, there is no risk of harm; the training has been delivered to children with and without disability in Cork and implies the use of voice, movement, and simple harmless musical tools.

The main benefit of this research will hopefully allow us to refine music training programmes that will impact positively on children with Down Syndrome or those that have other forms of cognitive impairments.

III. FURTHER DETAILS

Participation in voluntary. If you and your child decide to take part in the study and then change your mind, you and your child can withdraw from the study at any point and all their data will be permanently deleted. Behavioural, EEG and saliva/hair testing are independent components of the study, therefore, should one of them be distressing for a child, it can be dismissed. You and your child may also decide not to take part in this study, this will not affect your ability to take part in the classes.
The tests will be carried out by Dr. Eva McMullan, Dr. Jason S. Chan, Dr. Annalisa Setti, and Dr. Yvonne Nolan. Eva has completed a first aid course and a child protection course, and all investigators have obtained Garda Clearance. All information collected will not be associated with your child's name. When the results are published, it will be impossible to identify individual participants. All data will be securely stored on a password-protected computer, and any information about participant's identities will

be removed. The results will be presented in an MSc thesis prepared by Dr. McMullan. They will be seen by the supervisors (named above), an internal examiner and an external examiner in accordance with university regulations. The study may be read by future students and may be published in an academic journal. The data collected will remain on record for a period of at least 10 years. Only the investigators in this research project will have access to this material during this time. They are Dr. Jason S. Chan, Dr. Yvonne Nolan, Dr. Annalisa Setti and Dr. Eva McMullan-Glossop.

Section C AGREEMENT TO CONSENT

The research project and the treatment procedures associated with it have been fully explained to me. All experimental procedures have been identified, and no guarantee has been given regarding the possible results. I have had the opportunity to ask questions concerning any and all aspects of the project and any procedures involved. I am aware that participation is voluntary and that I may withdraw my consent for my child to participate at any time. I am aware that my decision not to participate or to withdraw will not restrict my child's access to music training normally available to me. The confidentiality of my records concerning my involvement in this project will be maintained in an appropriate manner.

I understand that the investigators have insurance as required by law in the event of injury resulting from this research.

I, the undersigned, hereby consent to my child's participation as a subject in the above-described project conducted at the School of Applied Psychology, Department of Music, and Department of Anatomy and Neuroscience.

I have received a copy of this consent form for my records. I understand that if I have any questions concerning this research, I can contact the researchers listed above. If I have further queries concerning my rights in connection with the research, I can contact the Clinical Research Ethics Committee of the Cork Teaching Hospitals, Lancaster Hall, 6 Little Hanover Street, Cork.

After reading the entire consent form, if you have no further questions about giving consent, please sign where indicated.

Investigator:

Name of Child

Name of Parent/Guardian:

O I consent for my child to begin the study.

O I do not consent; I do not wish my child to participate in the study

Please select the date

	Month	Day	Year
Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)





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University College Cork, Ireland
Coláiste na hOllscoile Corcaigh

School of
Applied Psychology

Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

DATA PROTECTION NOTICE

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

At University College Cork, we treat your privacy seriously. Any personal data which you provide to the University will be treated with the highest standards of security and confidentiality, in accordance with Irish and European Data Protection legislation. This notice sets out details of the information that we collect, how we process it and who we share it with. It also explains your rights under data protection law in relation to our processing of your data.

Who we are.

Throughout this Notice, "we", "us" and "our" refers to University College Cork, as study sponsor. For more information about us, please refer to our website: www.ucc.ie

How we will use your personal data

By participating in the study, information about your child (also called "personal data") will be collected for the study purposes mentioned in the Patient information leaflet. This personal data includes, for example:

- information that directly identifies your child (such as their name and their year of birth).
- their gender, ethnic and racial background.
- information on their health, medical condition, quality of life.
- information on their previous music education.

- information on their cognitive pre-tests and post-tests, including the Peabody Vocabulary Test.
- information contained in their hair and saliva samples and the results after analysis.

Personal data collected at any time during the study will be kept strictly confidential. To ensure confidentiality, the data generated during the study is **coded** with a number that will identify you in the study. Any information that leaves the clinical site will be labelled with your code instead of your name. Every person that has access to your uncoded data (that is kept at the School of Applied Psychology, University College Cork) is subject to professional secrecy and confidentiality.

Data that directly identifies you (uncoded data) is stored in your medical files at the clinical site in which the data have been obtained. The 'key' that connects the subject name and subject ID should be kept in a separate location to the data; securely (locked cupboard in a room with restricted access) by the investigator. All files which contain personally identifiable information (e.g., keys) should undergo an additional layer of encryption such as Microsoft Azure Information Protection

Unencrypted personal devices (laptop or phone) should not be used to acquire data.

Who will access my personal data?

Your uncoded data will only be accessible to the study investigator and study staff. Results of the study will be provided to the Clinical Research Ethics Committee of the Cork Teaching Hospitals (CREC) in compliance with national and international regulations on clinical studies.

The purpose and legal basis for collecting your data.

Any personal data you provide to us during the course of this study will be processed fairly and lawfully.

Signing the Informed Consent Form means that your personal data will be used for the purposes outlined in the Patient information leaflet (PIL).

Personal data collected during this study and the results of the study may be presented for scientific purposes. However, you will never be identified individually during these presentations. Your identity will not be revealed in any reports or publications.

The clinical site and the investigator will use your personal data within the scope defined above.

The General Data Protection Regulation allows us to process your data because you have provided your consent. You are entitled to withdraw your consent at any time.

How long we will keep your data?

The personal data collected in the study will be kept for a period of 10 years after the end of the study. However, your anonymised data will be held in accordance with FAIR data principles maybe stored indefinitely in an open data repository.

Your rights

You have various rights under data protection law, subject to certain exemptions, in connection with our processing of your personal data, including the right:

- to find out if we use your personal data, access your personal data, and receive copies of your personal data.
- to have inaccurate/incomplete information corrected and updated.
- in certain circumstances, to have your details deleted from systems that we use to process your personal data or have the use of your personal data restricted in certain ways.
- to object to certain processing of your data by UCC.
- to exercise your right to data portability where applicable (i.e. obtain a copy of your personal data in a commonly used electronic form.
- to withdraw your consent to the processing of your data up to 30 days of participation without giving a reason by notifying your decision to the investigator. This will not affect the lawfulness of processing data about you based on your consent before the withdrawal. If you withdraw your consent for data processing, your participation in the study stops and no further data will be collected from you. Your study physician will present you the options you have concerning your personal data.
- along with study withdrawal, you have the right to request the deletion of data about you, up to 30 days of participation, if your data are no longer necessary for the purposes of processing or there is no other legal ground for their further processing.
- after 30 days of participation, the link between your personal identity and your data will be destroyed, and we will no longer be able to identify you from the rest of the dataset. Thus, it will not be possible to withdraw your data.

If you wish to exercise any of these rights, please address your request to the study coordinator or the Information Compliance Manager University College Cork (details below).

Questions or Complaints

If you have any queries in relation to this study, please contact Dr. Eva McMullan at e.mcmullan@ucc.ie.

If you have any complaints in connection with our processing of your personal data, you can contact UCC's Information Compliance Manager: **Information Compliance**

Manager, Office of Corporate & Legal Affairs, University College Cork, Western Road, Cork E: foi@ucc.ie Tel: +353 21 4903949

You also have the right to lodge a complaint with the Data Protection Commission if you are unhappy with our processing of your personal data. Details of how to lodge a complaint can be found on the Data Protection Commission's website (www.dataprotection.ie) or by telephoning 1890 252 231.

Name of Parent/Guardian _____

Date _____

Name of Informant _____

Date _____

In submitting this form, you understand and agree that your personal details will be used by the School of Applied Psychology for the purposes outlined above.

Submit



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School of
Applied Psychology

Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

PARENT INFORMATION/CONSENT FORM FOR TYPICALLY DEVELOPING CHILDREN NOT ENGAGING WITH MUSIC

CONSENT BY SUBJECT FOR PARTICIPATION IN RESEARCH PROTOCOL

Section A

Protocol Number:_____ Participant Name:_____

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

Investigators Directing Research:
Chief Investigator: Dr. Jason S. Chan
Researchers: Dr Annalisa Setti
Dr. Yvonne Nolan
Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904530

Your child is being asked to participate in a research study that will assess how daily interaction with a specifically designed music programme will impact on the neurocognitive development of children with Down Syndrome and typically developing children. In order to decide whether or not you want to be a part of this research study, you should understand enough about its risks and benefits to make an informed judgment. This process is known as informed consent. This information and consent form gives detailed information about the research study, which will be

discussed with you. Once you understand the study, you will be asked to sign this form if you wish your child to participate.

Section B

I. WHAT YOU AND YOUR CHILD WILL BE ASKED TO DO?

If you agree that your child will participate in the study, he/she will be required to engage in various tests in two separate online testing sessions. The initial test will allow the investigators to mentally age-match the children using the Peabody Picture Vocabulary Test. This is an assessment tool for mental age widely used by clinicians and researchers; the test requires us to match words to pictures and cover our selected age range. It takes approximately half an hour online. The additional testing will include computer games.

Saliva testing will also be carried out, which will include collecting saliva samples at home. In addition, hair samples will be collected before the study begins. Both of these will be collected by the researcher at the start of the study and at the conclusion of the study all tests will be carried at the commencement of the study and once again after an eight-week period. An information evening will be held on Microsoft Teams in advance of the study to show you how to collect the hair and saliva samples. There is nothing your child needs to do between these testing sessions.

Parents will collect the saliva samples from their child at home. To do this you will be given 8 salivettes, 4 to be used at the start of the study and 4 to be used at the end of the study. These salivettes are plastic containers containing a swab. These have been age-appropriately designed for ease of collection from children. Samples will be collected immediately on waking, and then again at 15, 30 and 45 minutes after waking on one morning just before the start of the study and again one day at the end of the study. Levels of the hormones cortisol and oxytocin will be analysed in the laboratory from the saliva samples. The saliva sample must be stored at 4°C until collected by the researcher. Because cortisol accumulates in hair over time, we will also collect hair samples at the beginning and at the end of the study (a very small sample). A hair sample will be collected by you the parent using scissors as close to the scalp as possible at the first and last study visit. The sample will be wrapped in tin foil and stored at -20°C until collected by the researcher and cortisol concentration is measured in the lab.

You will be required to fill out two online questionnaires before the commencement of the study to establish basic details prior to the study. One questionnaire is entitled ‘Why Choose Music?’ that helps to establish details of your child’s prior engagement with music, if any and basic demographic information. In the second questionnaire, the ‘Paediatric Quality of Life Inventory, you are invited to answer questions to measure health-related quality of life prior to the study. You will be invited to answer the ‘Paediatric Quality of Life Inventory’ again after an eight-week period.

II. WHAT ARE THE POTENTIAL RISKS AND BENEFITS?

There is no risk of harm for children: the equipment used and the tests are non-harmful and have been traditionally used with various healthy and pathological groups in the literature; precaution will be taken to ensure that children are not excessively fatigued and distressed as a consequence of the length of the tests. All precautions have been taken to minimise risk. There will be the opportunity for breaks about every 5-7 minutes during the tests, but the testing can be paused at any point to ensure the comfort and well-being of the contributor.

The main benefit of this research will hopefully allow us to refine music training programmes that will impact positively on children with Down Syndrome or those that have other forms of cognitive impairments.

III. FURTHER DETAILS

Participation in voluntary. If you and your child decide to take part in the study and then changes your mind, you and your child can withdraw from the study at any point and all their data will be permanently deleted. Behavioural, EEG and saliva/hair testing are independent components of the study, therefore should one of them be distressing for a child, it can be dismissed. You and your child may also decide not to take part in this study, this will not affect your ability to take part in the classes.
The tests will be carried out by Dr. Eva McMullan, Dr. Jason S. Chan, Dr. Annalisa Setti, and Dr. Yvonne Nolan. Eva has completed a first aid course; a child protection course and all investigators have obtained Garda Clearance. All information collected will not be associated with your child's name. When the results are published it will be impossible to identify individual participants. All data will be securely stored on a password protected computer and any information about participant's identities will be removed. The results will be presented in an MSc thesis prepared by Dr. Eva McMullan. They will be seen by the supervisors (named above), an internal examiner and an external examiner in accordance with University Regulations. The study may be read by future students studying this course and may be published in an academic journal. The data collected will remain on record for a period of at least 10 years. Only the investigators in this research project will have access to this material during this time. They are Dr. Jason S. Chan, Dr. Yvonne Nolan, Dr. Annalisa Setti and Dr. Eva McMullan-Glossop.

Section C AGREEMENT TO CONSENT

The research project and the treatment procedures associated with it have been fully explained to me. All experimental procedures have been identified and no guarantee has been given about the possible results. I have had the opportunity to ask questions concerning any and all aspects of the project and any procedures involved. I am aware that participation is voluntary and that I may withdraw my consent for my child to participate at any time. I am aware that my decision not to participate or to withdraw will not restrict my child's access to musical training normally available to me.

Confidentiality of records concerning my involvement in this project will be maintained in an appropriate manner.

I understand that the investigators have such insurance as is required by law in the event of injury resulting from this research.

I, the undersigned, hereby consent my child to participate as a subject in the above-described project conducted at the School of Applied Psychology, Department of Music and Department of Anatomy and Neuroscience.

I have received a copy of this consent form for my records. I understand that if I have any questions concerning this research, I can contact the researchers listed above. If I have further queries concerning my rights in connection with the research, I can contact the Clinical Research Ethics Committee of the Cork Teaching Hospitals, Lancaster Hall, 6 Little Hanover Street, Cork.

After reading the entire consent form, if you have no further questions about giving consent, please sign where indicated.

Investigator:

Name of Child:

Signature of Parent/Guardian:

O I consent for my child to begin the study

O I do not consent; I do not wish my child to participate in the study

Please select the date

	Month	Day	Year
Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)





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Applied Psychology

Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

DATA PROTECTION NOTICE

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

At University College Cork, we treat your privacy seriously. Any personal data which you provide to the University will be treated with the highest standards of security and confidentiality, in accordance with Irish and European Data Protection legislation. This notice sets out details of the information that we collect, how we process it and who we share it with. It also explains your rights under data protection law in relation to our processing of your data.

Who we are.

Throughout this Notice, "we", "us" and "our" refers to University College Cork, as study sponsor. For more information about us, please refer to our website: www.ucc.ie

How we will use your personal data

By participating in the study, information about your child (also called "personal data") will be collected for the study purposes mentioned in the Patient information leaflet. This personal data includes, for example:

- information that directly identifies your child (such as their name and their year of birth).
- their gender, ethnic and racial background.
- information on their health, medical condition, quality of life.

- information on their previous music education.
- information on their cognitive pre-tests and post-tests, including the Peabody Vocabulary Test.
- information contained in their hair and saliva samples and the results after analysis.

Personal data collected at any time during the study will be kept strictly confidential. To ensure confidentiality, the data generated during the study is **coded** with a number that will identify you in the study. Any information that leaves the clinical site will be labelled with your code instead of your name. Every person that has access to your uncoded data (that is kept at the School of Applied Psychology, University College Cork) is subject to professional secrecy and confidentiality.

Data that directly identifies you (uncoded data) is stored in your medical files at the clinical site in which the data have been obtained. The 'key' that connects the subject name and subject ID should be kept in a separate location from the data; it should be securely stored (in a locked cupboard in a room with restricted access) by the investigator. All files that contain personally identifiable information (e.g., keys) should undergo an additional layer of encryption, such as Microsoft Azure Information Protection.

Unencrypted personal devices (laptops or phones) should not be used to acquire data.

Who will access my personal data?

Your uncoded data will only be accessible to the study investigator and staff. The study's results will be provided to the Clinical Research Ethics Committee of the Cork Teaching Hospitals (CREC) in compliance with national and international regulations on clinical studies.

The purpose and legal basis for collecting your data.

Any personal data you provide to us during the course of this study will be processed fairly and lawfully.

Signing the Informed Consent Form means that your personal data will be used for the purposes outlined in the Patient information leaflet (PIL).

Personal data collected during this study and the results of the study may be presented for scientific purposes. However, you will never be identified individually during these presentations. Your identity will not be revealed in any reports or publications.

The clinical site and the investigator will use your personal data within the scope defined above.

The General Data Protection Regulation allows us to process your data because you have provided your consent. You are entitled to withdraw your consent at any time.

How long we will keep your data?

The personal data collected in the study will be kept for 10 years after the end of the study. However, your anonymised data will be held in accordance with FAIR data principles and may be stored indefinitely in an open data repository.

Your rights

You have various rights under data protection law, subject to certain exemptions, in connection with our processing of your personal data, including the right:

- to find out if we use your personal data, access your personal data and receive copies of your personal data.
- to have inaccurate/incomplete information corrected and updated.
- in certain circumstances, to have your details deleted from systems that we use to process your personal data or have the use of your personal data restricted in certain ways.
- to object to certain processing of your data by UCC.
- to exercise your right to data portability where applicable (i.e. obtain a copy of your personal data in a commonly used electronic form.
- to withdraw your consent to the processing of your data up to 30 days of participation without giving a reason by notifying your decision to the investigator. This will not affect the lawfulness of processing data about you based on your consent before the withdrawal. If you withdraw your consent for data processing, your participation in the study stops and no further data will be collected from you. Your study physician will present you with the options you have concerning your personal data.
- along with study withdrawal, you have the right to request the deletion of data about you, up to 30 days of participation, if your data are no longer necessary for the purposes of processing or there is no other legal ground for their further processing.
- after 30 days of participation, the link between your personal identity and your data will be destroyed and we will no longer be able to identify you from the rest of the dataset, thus it will not be possible to withdraw your data.

If you wish to exercise any of these rights, please address your request to the study coordinator or the Information Compliance Manager at University College Cork (details below).

Questions or Complaints

If you have any queries in relation to this study, please contact Dr. Eva McMullan e.mcmullan@ucc.ie.

If you have any complaints in connection with our processing of your personal data, you can contact UCC's Information Compliance Manager: **Information Compliance**

Manager, Office of Corporate & Legal Affairs, University College Cork, Western Road, Cork E: foi@ucc.ie Tel: +353 21 4903949
You also have the right to lodge a complaint with the Data Protection Commission if you are unhappy with our processing of your personal data. Details of how to lodge a complaint can be found on the Data Protection Commission's website (www.dataprotection.ie) or by telephoning 1890 252 231.

Name of Parent/Guardian _____

Date _____

Name of Informant _____

Date _____

In submitting this form, you understand and agree that your personal details will be used by the School of Applied Psychology for the purposes outlined above.

Submit





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Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

CHILD INFORMATION/ASSENT FORM FOR CHILDREN WITH DOWN SYNDROME ENGAGING WITH MUSIC

CONSENT BY THE CHILD TO TAKE PART

Section A

Protocol Number:_____ Participant Name:_____

Name of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

Our names: Dr. Jason S. Chan, Dr Annalisa Setti, Dr. Yvonne Nolan, Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904530

You are asked to take part in a test that will assess how music will change the brains of kids with Down Syndrome and typically developing children.

We want to make sure you understand want you have to do before we start. You can decide if you want to take part or not. This is called informed consent. If you agree, we will ask you to sign this form before we start the study.

Section B

I. WHAT YOU WILL BE ASKED TO DO?

We want to know how the brain changes with music training and how this can be different with kids that have something called Down Syndrome. We want to know if music can help kids with Down Syndrome.

If you agree to take part, we will ask you to do the music program online every day for eight weeks, and you will need to come to your group music class online once a week. During this music programme you will get to sing songs and play some musical games and play instruments.

You will do a couple of online tests. These tests will be on the computer, some will be looking at and name some pictures and some will be moving things on a screen.

Your parents will take a saliva sample from your mouth, which just involves sucking on a piece of sponge attached to a stick, and also, they will take a small sample of hair.

We would like you to do these tests before the music training and after the music programme.

If you want to stop and no longer do this anymore, you just need to tell us. You will not get in any trouble, and you can still do the music classes.

The information will be held for a period of 10 years. Only the investigators in this research project will have access to this material during this time. Our names are Dr. Jason S. Chan, Dr. Yvonne Nolan, Dr. Annalisa Setti, and Dr. Eva McMullan-Glossop.

Section C AGREEMENT TO ASSENT

Do I know what I have to do? ☐

Did I get my questions answered? ☐

I know I can stop when I want, and I will not get in trouble. ☐

Investigator:

Name of Child

Name of Parent/Guardian:

O I agree to take part in this study.

O I do not agree to take part in this study.

Please select the date

	Month	Day	Year
Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)

Submit



CHILD INFORMATION/ASSENT FORM FOR CHILDREN WITH DOWN SYNDROME NOT ENGAGING WITH MUSIC

CONSENT BY THE CHILD TO TAKE PART

Section A

Protocol Number:_____ Participant Name:_____

Name of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

Our names: Dr. Jason S. Chan, Dr Annalisa Setti, Dr. Yvonne Nolan, Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904530

You are asked to take part in a test that will assess how music affects the brains of children with Down Syndrome and typically developing children.

We want to make sure you understand what you have to do before we start. You can decide if you want to take part or not. This is called informed consent. If you agree, we will ask you to sign this form before we start the study.

Section B

I. WHAT YOU WILL BE ASKED TO DO?

We want to know how the brain changes with music training and how this can be different with kids who have Down Syndrome. We want to know if music can help kids with Down Syndrome.

If you agree to take part, you will take some online tests. These tests will be all on the computer. Some will involve looking at and naming pictures, and some will involve moving things on the computer screen.

Your parents will take a saliva sample from your mouth, which just involves sucking on a piece of sponge attached to a stick, and they will take a small sample of hair.

We would like to you do these tests twice. One time and then again after about 8 weeks. You don't have to do anything in between.

If you want to stop and no longer do this anymore, you just need to tell us. You will not get in any trouble.

The information will be held for a period of at least 10 years. Only the investigators in this research project will have access to this material during this time. Our names are Dr. Jason S. Chan, Dr. Yvonne Nolan, Dr. Annalisa Setti, and Dr. Eva McMullan-Glossop.

Section C AGREEMENT TO ASSENT

Do I know what I have to do? ☐

Did I get my questions answered? ☐

I know I can stop when I want, and I will not get in trouble. ☐

Investigator:

Name of Child

Name of Parent/Guardian:

O I agree to take part in this study

O I do not agree to take part in this study

Please select the date

	Month	Day	Year

Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)

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University College Cork

Down Syndrome Cork

CHILD INFORMATION/ASSENT FORM FOR TYPICALLY DEVELOPING CHILDREN ENGAGING WITH MUSIC

CONSENT BY THE CHILD TO TAKE PART

Section A

Protocol Number:_____ Participant Name:

Name of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

Our names: Dr. Jason S. Chan, Dr Annalisa Setti, Dr. Yvonne Nolan, Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904530

You are asked to take part in a test that will assess how music will change the brains of kids with Down Syndrome and typically developing children.

We want to make sure you understand want you have to do before we start. You can decide if you want to take part or not. This is called informed consent. If you agree, we will ask you to sign this form before we start the study.

Section B

I. WHAT YOU WILL BE ASKED TO DO?

We want to know how the brain changes with music training and how this can be different with kids that have something called Down Syndrome. We want to know if music can help kids with Down Syndrome.

If you agree to take part, we will ask you to do the music program online every day for eight weeks, and you will need to come to your group music class online once a week. During this music programme you will get to sing songs and play some musical games and play instruments.

You will do a couple of online tests. These tests will be on the computer. Some will involve looking at and naming pictures, and some will involve moving things on a screen.

Your parents will take a saliva sample from your mouth which just involves sucking on a piece of sponge attached to a stick and, they will take a small sample of hair.

We would like you to do these tests before the music training and after the music programme.

If you want to stop doing this, you just need to tell us. You will not get in any trouble, and you can still take the music classes.

The information will be held for a period of 10 years. Only the investigators in this research project will have access to this material during this time. Our names are Dr. Jason S. Chan, Dr. Yvonne Nolan, Dr. Annalisa Setti and Dr. Eva McMullan-Glossop.

Section C AGREEMENT TO ASSENT

Do I know what I have to do? ☐

Did I get my questions answered? ☐

I know I can stop when I want, and I will not get in trouble. ☐

Investigator:

Name of Child

Name of Parent/Guardian:

O I agree to take part in this study.

O I do not agree to take part in this study.

Please select the date

	Month	Day	Year
Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)

Submit



CHILD INFORMATION/ASSENT FORM FOR TYPICALLY DEVELOPING CHILDREN NOT ENGAGING WITH MUSIC

CONSENT BY THE CHILD TO TAKE PART

Section A

Protocol Number:_____ Participant Name:_____

Name of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

Our names: Dr. Jason S. Chan, Dr Annalisa Setti, Dr. Yvonne Nolan, Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904530

You are asked to take part in a test that will assess how music affects the brains of children with Down Syndrome and typically developing children.

We want to make sure you understand what you have to do before we start. You can decide if you want to take part or not. This is called informed consent. If you agree, we will ask you to sign this form before we start the study.

Section B

I. WHAT YOU WILL BE ASKED TO DO?

We want to know how the brain changes with music training and how this can be different with kids who have Down Syndrome. We want to know if music can help kids with Down Syndrome.

If you agree to take part, you will take some online tests. These tests will be all on the computer. Some will involve looking at and naming pictures, and some will involve moving things on the computer screen.

Your parents will take a saliva sample from your mouth, which just involves sucking on a piece of sponge attached to a stick, and they will take a small sample of hair.

We would like to you do these tests twice. One time and then again after about 8 weeks. You don't have to do anything in between.

If you want to stop and no longer do this anymore, you just need to tell us. You will not get in any trouble.

The information will be held for a period of at least 10 years. Only the investigators in this research project will have access to this material during this time. Our names are Dr. Jason S. Chan, Dr. Yvonne Nolan, Dr. Annalisa Setti, and Dr. Eva McMullan-Glossop.

Section C AGREEMENT TO ASSENT

Do I know what I have to do? ☐

Did I get my questions answered? ☐

I know I can stop when I want, and I will not get in trouble. ☐

Investigator:

Name of Child

Name of Parent/Guardian:

O I agree to take part in this study

O I do not agree to take part in this study

Please select the date

	Month	Day	Year

Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)

Submit





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Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

INFORMATION LEAFLET AND INFORMED ASSENT FORM FOR CHILDREN WITH DOWN SYNDROME ENGAGING WITH MUSIC TRAINING

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

Chief Investigator: Dr. Jason Chan (UCC)

My name is Eva, and my job is to research and test your brain to see how important music is in how we grow. We want to know how your brain works and the brain of other children. I am going to give you information and invite you to be part of a research study. You can choose whether or not you want to take part.

What is a study?

A research study is what you do when you want to learn about something or find out something new.

Why is this study being done?

We are doing this study with music to find out how it can help make our brains healthier.

What will happen to me if I take part? I will explain what happens to you, the games and the tests that we will do. If you can, you will be asked to write your name on a form.

This form is to say that you understand the study and what will happen. You will be given your own copy of the form to keep, as well as this leaflet.

This is what you will have to do if you say yes:

You will get to take part in an online music class every Saturday morning and at home watch some music videos. You will get to play musical instruments and play some

music games. You will get to look at some pictures and tell us what you see. You will be asked to play some games on the computer. This is to give your brain some exercise. When you are at home, we will ask you if you can give us some saliva and hair that your mum will take.

Do I have to say yes?

No – not at all. It's up to you! Just say if you don't want to join in. Nobody will mind. If you change your mind, that's okay as well; if you want to stop, you can say it, and it is no problem. This will not change the way you learn music.

What shall I do now?

Now you know about the study you need to think about if you want to take part in the study.

Who can I ask about this?

Your mum or dad (or carer) have been given lots of information, but if you have any questions or would like to speak to me or the music testing team, you can call.
Eva and Jason on 021-4904530

Will the study upset or help me?

No - the study will not upset you. We hope that the information we get from this study will encourage boys and girls to play music in the future.

Thank you very much for taking time to read this.
Please ask any questions if you need to.



INFORMED ASSENT FORM FOR CHILDREN WITH DOWN SYNDROME ENGAGING WITH MUSIC TRAINING

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

To be completed by the child and their parent/guardian

Please highlight all you agree with:
(Child)

Has somebody explained this study to you?	**Yes / No**
Do you understand what the study is about?	**Yes / No**
Have you asked all the questions you want?	**Yes / No**
Have you had your questions answered in a way you understand?	**Yes / No**
Do you understand it's OK to stop taking part at any time?	**Yes / No**
Are you happy to take part?	**Yes / No**

If any answers are 'no' or you **don't** want to take part, **don't** sign your name! If you do want to take part, please write your name and today's date.

Your Name: _____

Today's Date: _____

Your parent or guardian must write their name here too if they are happy for you to do the study.

Parent/Guardian Full Name: _____

Today's Date: _____

The researcher who explained this study to you needs to sign too:

Researcher Full Name: _____

Today's Date: _____

When completed, 1 (original) is to be kept in the researcher site file, and 1 (copy) is to be given to the participant





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University College Cork

Down Syndrome Cork

INFORMATION LEAFLET AND INFORMED ASSENT FORM FOR CHILDREN WITH DOWN SYNDROME THAT ARE NOT ENGAGING WITH MUSIC TRAINING

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

Chief Investigator: Dr. Jason Chan (UCC)

My name is Eva, and my job is to research and test your brain to see how amazing it is. We want to know how your brain works and the brains of other children. I am going to give you information and invite you to be part of a research study. You can choose whether or not you want to take part.

What is a study?

A research study is what you do when you want to learn about something or find out something new.

Why is this study being done?

We are doing this study to find how we can help make our brains healthier.

What will happen to me if I take part? I will explain what happens to you, the games and tests that we will do. If you can, you will be asked to write your name on a form.

This form is to say that you understand the study and what will happen. You will be given your own copy of the form to keep, as well as this leaflet.

This is what you will have to do if you say yes:

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No – not at all. It's up to you! Just say if you don't want to join in. Nobody will mind. If you change your mind, that's okay as well. If you want to stop, you can say it, and it is no problem.

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Your mum or dad (or carer) have been given lots of information, but if you have any questions or would like to speak to me or the testing team, you can call Eva and Jason on 021-4904530

Will the study upset or help me?

No - the study will not upset you. We hope that the information we get from this study will encourage boys and girls to play music in the future.

Thank you very much for taking time to read this. Please ask any questions if you need to.



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<u>INFORMED ASSENT FORM FOR CHILDREN WITH DOWN SYNDROME THAT ARE NOT ENGAGING WITH MUSIC TRAINING</u>

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

To be completed by the child and their parent/guardian

Please highlight all you agree with:
(Child)

Has somebody explained this study to you?	**Yes / No**
Do you understand what the study is about?	**Yes / No**
Have you asked all the questions you want?	**Yes / No**
Have you had your questions answered in a way you understand?	**Yes / No**
Do you understand it's OK to stop taking part at any time?	**Yes / No**
Are you happy to take part?	**Yes / No**

If <u>any</u> answers are 'no' or you **don't** want to take part, **don't** sign your name! If you <u>do</u> want to take part, please write your name and today's date.

Your Name: _____

Today's Date: _____

Your parent or guardian must write their name here too if they are happy for you to do the study.

Parent/Guardian Full Name: _____

Today's Date: _____

The researcher who explained this study to you needs to sign too:

Researcher Full Name: _____

Today's Date: _____

When completed, 1 (original) to be kept in researcher site file and 1 (copy) to be given to the participant





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INFORMATION LEAFLET AND INFORMED ASSENT FORM FOR TYPICALLY DEVELOPING CHILDREN ENGAGING WITH MUSIC TRAINING

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

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What shall I do now?

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Who can I ask about this?

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INFORMED ASSENT FORM FOR TYPICALLY DEVELOPING CHILDREN ENGAGING WITH MUSIC TRAINING

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

To be completed by the child and their parent/guardian

Please highlight all you agree with:
(Child)

Has somebody explained this study to you?	**Yes / No**
Do you understand what the study is about?	**Yes / No**
Have you asked all the questions you want?	**Yes / No**
Have you had your questions answered in a way you understand?	**Yes / No**
Do you understand it's OK to stop taking part at any time?	**Yes / No**
Are you happy to take part?	**Yes / No**

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Your Name: _____

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Parent/Guardian Full Name: _____

Today's Date: _____

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INFORMATION LEAFLET AND INFORMED ASSENT FORM FOR TYPICALLY DEVELOPING CHILDREN THAT ARE NOT ENGAGING WITH MUSIC TRAINING

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

Chief Investigator: Dr. Jason Chan (UCC)

My name is Eva, and my job is to research and test your brain to see how amazing it is. We want to know how your brain works and the brains of other children. I am going to give you information and invite you to be part of a research study. You can choose whether or not you want to take part.

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INFORMED ASSENT FORM FOR TYPICALLY DEVELOPING CHILDREN THAT ARE NOT ENGAGING WITH MUSIC TRAINING

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

To be completed by the child and their parent/guardian

Please highlight all you agree with:
(Child)

Has somebody explained this study to you?	**Yes / No**
Do you understand what the study is about?	**Yes / No**
Have you asked all the questions you want?	**Yes / No**
Have you had your questions answered in a way you understand?	**Yes / No**
Do you understand it's OK to stop taking part at any time?	**Yes / No**
Are you happy to take part?	**Yes / No**

If any answers are 'no' or you **don't** want to take part, **don't** sign your name! If you do want to take part, please write your name and today's date.

Your Name: _____

Today's Date: _____

Your parent or guardian must write their name here too if they are happy for you to do the study.

Parent/Guardian Full Name: _____

Today's Date: _____

The researcher who explained this study to you needs to sign too:

Researcher Full Name: _____

Today's Date: _____

When completed, 1 (original) to be kept in researcher site file and 1 (copy) to be given to the participant





UCC University College Cork, Ireland Coláiste na hOllscoile Corcaigh | School of Applied Psychology | Down Syndrome Cork | Department of Anatomy & Neuroscience University College Cork

INFORMATION LEAFLET

Title of Study: Investigation of the neurological effects of musical training in children with Down Syndrome

INFORMATION LEAFLET FOR PARENTS OF CHILDREN WHO HAVE ALREADY TAKEN PART IN THE STUDY

Because of your involvement in the study thus far, you are now invited to fill out an additional questionnaire as part of the current study. Entitled 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention,' this Questionnaire has been designed to give you, the parent, the opportunity to reflect and share your observations during and after the intervention.

WHAT IS INVOLVED?

In advance of filling out the questionnaire, you will be asked to give your consent, which will be distributed via a Qualtrics link to the questionnaire. This Questionnaire will include open and closed questions that will permit you, the parent, to input your noted observations during and after the intervention. You will be asked to enter your Subject ID to retain anonymity.

BENEFITS

As researchers, we recognise the vital role that parental observations of their children have in assessing the intervention. Your comments and the neurological and behavioural measures will help comprehensively evaluate the programme.





For Further Information

Contact: Dr. Eva McMullan-Glossop e.mcmullan@ucc.ie or jason.chan@ucc.ie ***or***
Tel: 021 494536

Celebrating Music Learning through fun and games by taking inspiration from children themselves.



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PARENT INFORMATION/CONSENT FORM FOR CHILDREN WITH DOWN SYNDROME ENGAGING WITH MUSIC

CONSENT BY PARENT FOR CONTRIBUTION TO 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention'

Section A

Protocol Number:_____ Participant Name:_____

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

Investigators Directing Research:

Chief Investigator: Dr. Jason S. Chan,
Researchers: Dr Annalisa Setti,
Dr. Yvonne Nolan,
Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904520

What is Being Asked

You are being asked to fill out an additional questionnaire as part of the existing project, 'An Investigation of the Neurological Effects of Musical Training in children with Down Syndrome. The questionnaire is entitled 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention' and will be completed after the intervention. You will be asked both open and closed questions.

Risks and Benefits

The are no risks in taking part in this Questionnaire. The main benefit of this research will be to provide us with the knowledge to refine music training programmes that will positively impact children with Down Syndrome or those that have other forms of cognitive impairments.

Section B AGREEMENT TO CONSENT

I understand all that is being asked in relation to my participation in the above survey.

I, the undersigned, hereby consent to participate as a subject in the above-described Questionnaire conducted at the School of Applied Psychology, Department of Music, and Department of Anatomy and Neuroscience.

I have received a copy of this consent form for my records. I understand that if I have any questions concerning this research, I can contact the researchers listed above. If I have further queries concerning my rights in connection with the research, I can contact the Clinical Research Ethics Committee of the Cork Teaching Hospitals, Lancaster Hall, 6 Little Hanover Street, Cork.

After reading the entire consent form, if you have no further questions about giving consent, please sign where indicated.

Name of Parent/Guardian_____

O I consent to take part in this Questionnaire.

O I do not consent; I do not wish to take part in this Questionnaire.

Please select the date

	Month	Day	Year
Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)





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Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

DATA PROTECTION NOTICE

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

At University College Cork, we treat your privacy seriously. Any personal data which you provide to the University will be treated with the highest standards of security and confidentiality, in accordance with Irish and European Data Protection legislation. This notice sets out details of the information that we collect, how we process it and who we share it with. It also explains your rights under data protection law in relation to our processing of your data.

Who we are.

Throughout this Notice, "we", "us" and "our" refers to University College Cork, as study sponsor. For more information about us, please refer to our website: www.ucc.ie

How we will use your personal data

By participating in the study, information about your child (also called "personal data") will be collected for the study purposes mentioned in the Patient information leaflet. This personal data includes, for example:

- information that directly identifies your child (such as their name and their year of birth).
- their gender, ethnic and racial background.
- information on their health, medical condition, quality of life.
- information on their previous music education.
- information on their cognitive pre-tests and post-tests, including the Peabody Vocabulary Test.
- information contained in their hair and saliva samples and the results after analysis.

Personal data collected at any time during the study will be kept strictly confidential. To ensure confidentiality, the data generated during the study is **coded** with a number that will identify you in the study. Any information that leaves the clinical site will be labelled with your code instead of your name. Every person that has access to your uncoded data (that is kept at the School of Applied Psychology, University College Cork) is subject to professional secrecy and confidentiality.

Data that directly identifies you (uncoded data) is stored in your medical files at the clinical site in which the data have been obtained. The 'key' that connects the subject name and subject ID should be kept in a separate location from the data; it should be securely stored (in a locked cupboard in a room with restricted access) by the investigator. All files that contain personally identifiable information (e.g., keys) should undergo an additional layer of encryption, such as Microsoft Azure Information Protection.

Unencrypted personal devices (laptops or phones) should not be used to acquire data.

Who will access my personal data?

Your uncoded data will only be accessible to the study investigator and staff. The study's results will be provided to the Clinical Research Ethics Committee of the Cork Teaching Hospitals (CREC) in compliance with national and international regulations on clinical studies.

The purpose and legal basis for collecting your data.

Any personal data you provide to us during the course of this study will be processed fairly and lawfully.

Signing the Informed Consent Form means that your personal data will be used for the purposes outlined in the Patient information leaflet (PIL).

Personal data collected during this study and the results of the study may be presented for scientific purposes. However, you will never be identified individually during these presentations. Your identity will not be revealed in any reports or publications.

The clinical site and the investigator will use your personal data within the scope defined above.

The General Data Protection Regulation allows us to process your data because you have provided your consent. You are entitled to withdraw your consent at any time.

How long we will keep your data?

The personal data collected in the study will be kept for 10 years after the end of the study. However, your anonymised data will be held in accordance with FAIR data principles and may be stored indefinitely in an open data repository.

Your rights

You have various rights under data protection law, subject to certain exemptions, in connection with our processing of your personal data, including the right:

- to find out if we use your personal data, access your personal data and receive copies of your personal data.
- to have inaccurate/incomplete information corrected and updated.
- in certain circumstances, to have your details deleted from systems that we use to process your personal data or have the use of your personal data restricted in certain ways.
- to object to certain processing of your data by UCC.
- to exercise your right to data portability where applicable (i.e. obtain a copy of your personal data in a commonly used electronic form.
- to withdraw your consent to the processing of your data up to 30 days of participation without giving a reason by notifying your decision to the investigator. This will not affect the lawfulness of processing data about you based on your consent before the withdrawal. If you withdraw your consent for data processing, your participation in the study stops and no further data will be collected from you. Your study physician will present you with the options you have concerning your personal data.
- along with study withdrawal, you have the right to request the deletion of data about you, up to 30 days of participation, if your data are no longer necessary for the purposes of processing or there is no other legal ground for their further processing.
- after 30 days of participation, the link between your personal identity and your data will be destroyed and we will no longer be able to identify you from the rest of the dataset, thus it will not be possible to withdraw your data.

If you wish to exercise any of these rights, please address your request to the study coordinator or the Information Compliance Manager at University College Cork (details below).

Questions or Complaints

If you have any queries in relation to this study, please contact Dr. Eva McMullan e.mcmullan@ucc.ie.

If you have any complaints in connection with our processing of your personal data, you can contact UCC's Information Compliance Manager: **Information Compliance Manager, Office of Corporate & Legal Affairs, University College Cork, Western Road, Cork E: foi@ucc.ie Tel: +353 21 4903949**
You also have the right to lodge a complaint with the Data Protection Commission if you are unhappy with our processing of your personal data. Details of how to lodge a complaint can be found on the Data Protection Commission's website (www.dataprotection.ie) or by telephoning 1890 252 231.

Name of Parent/Guardian _____

Date _____

Name of Informant _____

Date _____

In submitting this form, you understand and agree that your personal details will be used by the School of Applied Psychology for the purposes outlined above.

Submit





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Down Syndrome Cork

PARENT INFORMATION/CONSENT FORM FOR TYPICALLY DEVELOPING CHILDREN ENGAGING WITH MUSIC

CONSENT BY PARENT FOR CONTRIBUTION TO 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention'

Section A

Protocol Number:_____ Participant Name:_____

Title of Protocol: Investigation of the neurological effects of musical training in children with Down Syndrome

Investigators Directing Research:

Chief Investigator: Dr. Jason S. Chan,
Researchers: Dr Annalisa Setti,
Dr. Yvonne Nolan,
Dr. Eva McMullan-Glossop

Contact information: jason.chan@ucc.ie and e.mcmullan@ucc.ie; Tel: 021-4904520

What is Being Asked

You are being asked to fill out an additional questionnaire as part of the existing project, 'An Investigation of the Neurological Effects of Musical Training in children with Down Syndrome. The questionnaire is entitled 'A Qualitative Parental Description of Your Child's Experience of the Music Intervention' and will be completed after the intervention. You will be asked both open and closed questions.

Risks and Benefits

The are no risks in taking part in this Questionnaire. The main benefit of this research will be to provide us with the knowledge to refine music training programmes that will positively impact children with Down Syndrome or those that have other forms of cognitive impairments.

Section B AGREEMENT TO CONSENT

I understand all that is being asked in relation to my participation in the above survey.

I, the undersigned, hereby consent to participate as a subject in the above-described Questionnaire conducted at the School of Applied Psychology, Department of Music, and Department of Anatomy and Neuroscience.

I have received a copy of this consent form for my records. I understand that if I have any questions concerning this research, I can contact the researchers listed above. If I have further queries concerning my rights in connection with the research, I can contact the Clinical Research Ethics Committee of the Cork Teaching Hospitals, Lancaster Hall, 6 Little Hanover Street, Cork.

After reading the entire consent form, if you have no further questions about giving consent, please sign where indicated.

Name of Parent/Guardian_____

O I consent to take part in this Questionnaire.

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	Month	Day	Year
Please Select:	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)



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Unencrypted personal devices (laptops or phones) should not be used to acquire data.

Who will access my personal data?

Your uncoded data will only be accessible to the study investigator and staff. The study's results will be provided to the Clinical Research Ethics Committee of the Cork Teaching Hospitals (CREC) in compliance with national and international regulations on clinical studies.

The purpose and legal basis for collecting your data.

Any personal data you provide to us during the course of this study will be processed fairly and lawfully.

Signing the Informed Consent Form means that your personal data will be used for the purposes outlined in the Patient information leaflet (PIL).

Personal data collected during this study and the results of the study may be presented for scientific purposes. However, you will never be identified individually during these presentations. Your identity will not be revealed in any reports or publications.

The clinical site and the investigator will use your personal data within the scope defined above.

The General Data Protection Regulation allows us to process your data because you have provided your consent. You are entitled to withdraw your consent at any time.

How long we will keep your data?

The personal data collected in the study will be kept for 10 years after the end of the study. However, your anonymised data will be held in accordance with FAIR data principles and may be stored indefinitely in an open data repository.

Your rights

You have various rights under data protection law, subject to certain exemptions, in connection with our processing of your personal data, including the right:

- to find out if we use your personal data, access your personal data and receive copies of your personal data.
- to have inaccurate/incomplete information corrected and updated.
- in certain circumstances, to have your details deleted from systems that we use to process your personal data or have the use of your personal data restricted in certain ways.
- to object to certain processing of your data by UCC.
- to exercise your right to data portability where applicable (i.e. obtain a copy of your personal data in a commonly used electronic form.
- to withdraw your consent to the processing of your data up to 30 days of participation without giving a reason by notifying your decision to the investigator. This will not affect the lawfulness of processing data about you based on your consent before the withdrawal. If you withdraw your consent for data processing, your participation in the study stops and no further data will be collected from you. Your study physician will present you with the options you have concerning your personal data.
- along with study withdrawal, you have the right to request the deletion of data about you, up to 30 days of participation, if your data are no longer necessary for the purposes of processing or there is no other legal ground for their further processing.
- after 30 days of participation, the link between your personal identity and your data will be destroyed and we will no longer be able to identify you from the rest of the dataset, thus it will not be possible to withdraw your data.

If you wish to exercise any of these rights, please address your request to the study coordinator or the Information Compliance Manager at University College Cork (details below).

Questions or Complaints

If you have any queries in relation to this study, please contact Dr. Eva McMullan e.mcmullan@ucc.ie.

If you have any complaints in connection with our processing of your personal data, you can contact UCC's Information Compliance Manager: **Information Compliance Manager, Office of Corporate & Legal Affairs, University College Cork, Western Road, Cork E: foi@ucc.ie Tel: +353 21 4903949**
You also have the right to lodge a complaint with the Data Protection Commission if you are unhappy with our processing of your personal data. Details of how to lodge a complaint can be found on the Data Protection Commission's website (www.dataprotection.ie) or by telephoning 1890 252 231.

Name of Parent/Guardian _____

Date _____

Name of Informant _____

Date _____

In submitting this form, you understand and agree that your personal details will be used by the School of Applied Psychology for the purposes outlined above.

Submit



Appendix B: Questionnaires



UCC
University College Cork, Ireland
Coláiste na hOllscoile Corcaigh

School of
Applied Psychology

Department of Anatomy & Neuroscience
University College Cork

Down Syndrome Cork

WHY CHOOSE MUSIC? *

This questionnaire, for the study entitled 'Investigation of the Neurological effects of Musical Training in Children with Down Syndrome', aims to gather important information about you and your child's engagement with music. The survey includes the following sections:

- **Section One:** **Background Information**
- **Section Two:** **Rate Your Own Engagement with Music**
- **Section Three** **Engaged with Music4Children**
- **Section Four** **Not Engaged with Music**
- **Section Five:** **Open Ended Questions**

Section One: Background Information

What is your sex? male ☐ female ☐

Please specify your age category. 18-2 ☐ 30-49 ☐ 50+ ☐

What is your academic qualification?

_____
_____

Is your child currently/has been engaged with a Music4Children programme?

_____
_____

Section Two: Please Rate your own engagement with Music, with 0 being not true of me and 7 being very true of me.

1. **I listen to music regularly.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

2. **I regularly attend concerts.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

3. **I play one or more musical instruments.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

4. **I am myself a musician.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

5. **I have been paid to teach or perform music.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

Section Three is for those ***engaged*** with Music4Children. If your child is not ***engaged*** in the Music4Children programme, please proceed to ***Section Four***.

Section Three: Engaged with Music4Children

1. **My child is involved in music because I wanted to have a variety of activities.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

2. **My child is engaged with a music programme as I feel it would improve their musical skill.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

3. **My child is participating in music because their friends are doing music.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

4. **My child asked to be involved in music.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

5. **I thought being involved with music would improve their social skills.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

6. **I feel that music is an important experience for all children.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

7. **Please state any other reasons or comments as to why your child is engaged with music.**

_____

_____

_____

After ***Section Three*** is completed, please move to ***Section Five***

Section Four: Not Engaged with Music

1. **My child has never engaged with music because they refused to attend classes.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

2. **I do not think music is a valuable activity at this time in my child's life.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

3. **I do not think that he/she would enjoy it.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

4. **I do not think that they have any musical talent.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

5. **They never showed any interest in musical activity.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

6. **I was not aware of any programmes in our area.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

7. **Music lessons are too expensive.**

Not at all true of me Very true of me

0 1 2 3 4 5 6 7

Section Five: Open Ended Questions

1. **Do you feel that the reasons for involving your other child/children in music programmes were any different from those of the child discussed?**

_____
_____
_____
_____

2. **How important do you think music is in a child's early experiences?**

_____
_____
_____
_____

3. **Please share any other thoughts or comments**

Thank You, You Have now Completed the Questionnaire

*Adapted from 'Why Choose Music' from Paterson, J. (2008). *Why choose music? A study in parental motivation toward formal music learning and the cultural beliefs regarding its benefits* Sydney eScholarship. http://hdl.handle.net/2123/3995

ID# _____

Date _____

PedsQL™

Paediatric Quality of Life Inventory

Version 4.0 - English (Ireland)

PARENT REPORT for TODDLERS (ages 2-4)

INSTRUCTIONS

On the following page is a list of things that might be a problem for your chlid. Please tell us how much of a problem each one has been for your child during the past ONE month by circling:

0 if it is never a problem
1 if it is almost never a problem
2 if it is sometimes a problem
3 if it is often a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

Paediatric Quality of Life Inventory for 2–4-year-olds. Reproduced with permission from Prof. James Varni © (Varni et al. 2001).

*In the past **ONE month**, how much of a **problem** has your child had with...*

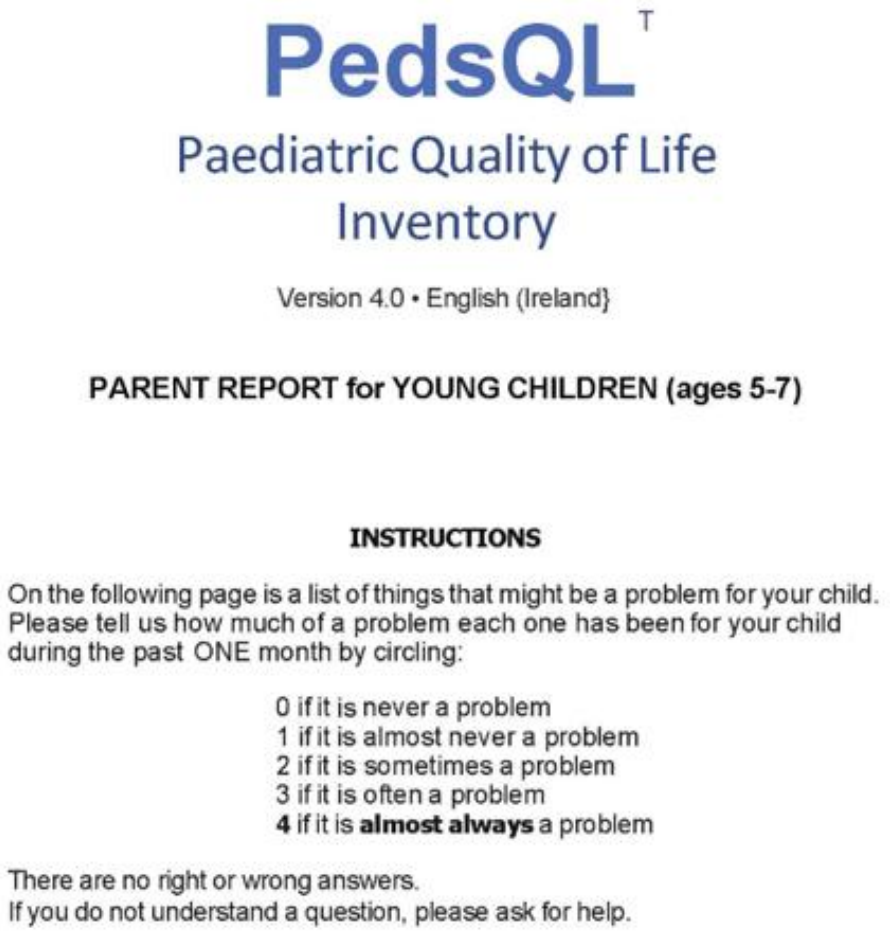
PHYSICAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in active play or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Bathing	0	1	2	3	4
6. Helping to pick up his or her toys	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Low energy level	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or down	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Having trouble sleeping	0	1	2	3	4
5. Worrying	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Playing with other children	0	1	2	3	4
2. Other children not wanting to play with him or her	0	1	2	3	4
3. Getting teased by other children	0	1	2	3	4
4. Not able to do things that other children his or her age can do	0	1	2	3	4
5. Keeping up when playing with other children	0	1	2	3	4

** Please complete this section if your child attends school or daycare*

SCHOOL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Doing the same school activities as peers	0	1	2	3	4
2. Missing school/daycare because of not feeling well	0	1	2	3	4
3. Missing school/daycare to go to the doctor or hospital	0	1	2	3	4



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PARENT REPORT for YOUNG CHILDREN (ages 5-7)

INSTRUCTIONS

On the following page is a list of things that might be a problem for your child. Please tell us how much of a problem each one has been for your child during the past ONE month by circling:

0 if it is never a problem
1 if it is almost never a problem
2 if it is sometimes a problem
3 if it is often a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

(Paediatric Quality of Life Inventory for 5–7-year-olds. Reproduced with permission from Prof. James Varni © Varni et al., 2001).

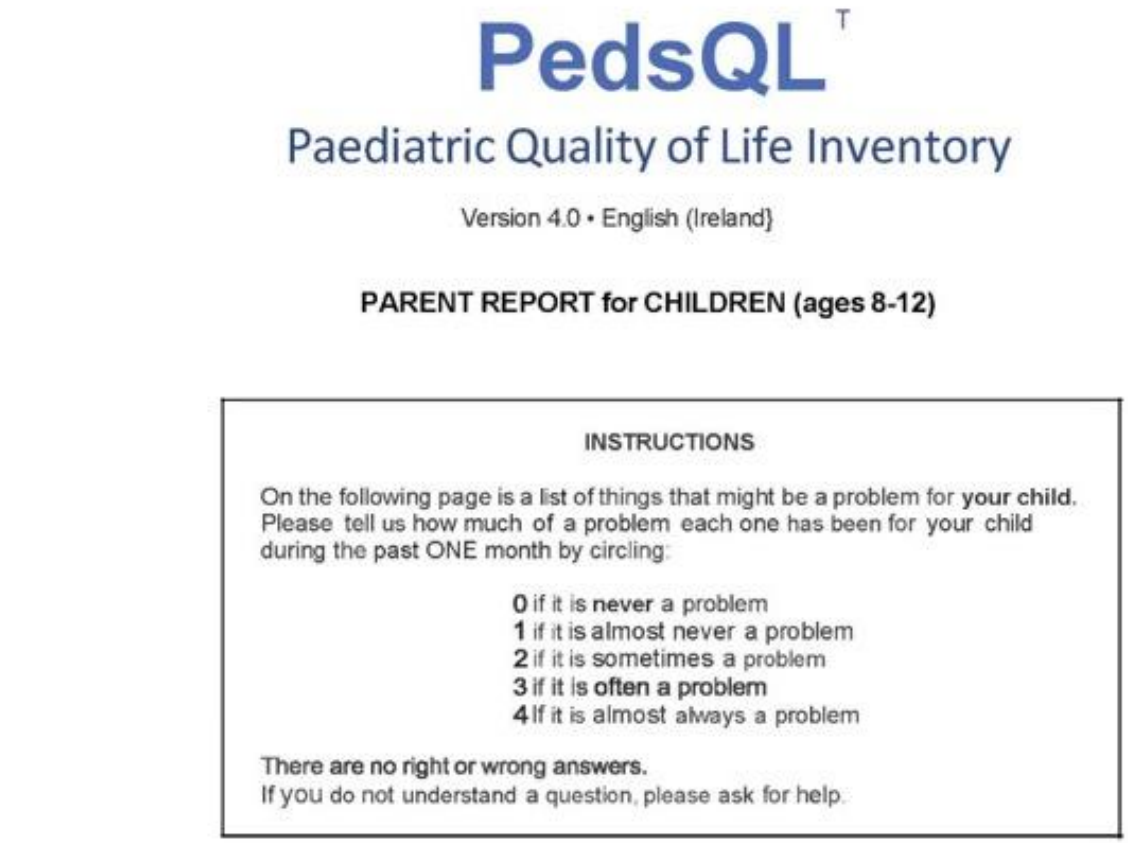
*In the past **ONE month**, how much of a **problem** has your child had with.*

PHYSICAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking more than 100 meters	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports activities or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Taking a bath or shower by him or herself	0	1	2	3	4
6. Doing chores, like picking up his or her toys	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8. Low energy level	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or down	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Having trouble sleeping	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Getting along with other children	0	1	2	3	4
2. Other children not wanting to be his or her friend	0	1	2	3	4
3. Getting teased by other children	0	1	2	3	4
4. Not able to do things that other children his or her age can do	0	1	2	3	4
5. Keeping up when playing with other children	0	1	2	3	4

SCHOOL FUNCTIONING *(problems with...)*	Never	Almost Never	some-times	Often	Almost Always
1. Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with school activities	0	1	2	3	4
4. Missing school because of not feeling well	0	1	2	3	4
5. Missing school to go to the doctor or hospital	0	1	2	3	4



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Paediatric Quality of Life Inventory

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PARENT REPORT for CHILDREN (ages 8-12)

INSTRUCTIONS

On the following page is a list of things that might be a problem for **your child**. Please tell us how much of a problem each one has been for your child during the past ONE month by circling:

0 if it is **never** a problem
1 if it is almost never a problem
2 if it is sometimes a problem
3 if it is **often a problem**
4 If it is almost always a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

Paediatric Quality of Life Inventory for 8-12-year-olds. Reproduced with permission from Prof. James Varni © Varni et al., 2001).

*In the past **ONE month**, how much of a **problem** has your child had with...*

PHYSICAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Walking more than 100 meters	0	1	2	3	4
2. Running	0	1	2	3	4
3. Participating in sports activities or exercise	0	1	2	3	4
4. Lifting something heavy	0	1	2	3	4
5. Taking a bath or shower by him or herself	0	1	2	3	4
6. Doing chores around the house	0	1	2	3	4
7. Having hurts or aches	0	1	2	3	4
8 Low energy level	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Feeling afraid or scared	0	1	2	3	4
2. Feeling sad or down	0	1	2	3	4
3. Feeling angry	0	1	2	3	4
4. Having trouble sleeping	0	1	2	3	4
5. Worrying about what will happen to him or her	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with...)*	Never	Almost Never	Some-times	Often	Almost Always
1. Getting along with other children	0	1	2	3	4
2. Other children not wanting to be his or her friend	0	1	2	3	4
3. Getting teased by other children	0	1	2	3	4
4. Not able to do things that other children his or her age can do	0	1	2	3	4
5. Keeping up when playing with other children	0	1	2	3	4

WORK/STUDIES FUNCTIONING *(problems with.. .)*	Never	Almost Never	Some-times	Often	Almost Always
1. Paying attention in class	0	1	2	3	4
2. Forgetting things	0	1	2	3	4
3. Keeping up with schoolwork	0	1	2	3	4
4. Missing school because of not feeling well	0	1	2	3	4
5. Missing school to go to the doctor or hospital	0	1	2	3	4



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University College Cork

Down Syndrome Cork

A Qualitative Parental Description of Your Child's Experience of the Music Intervention

This form should be completed by the parent/guardian of the child who engaged with the music intervention.

Q 1 Subject ID _____

Q 2 What aspects of the music intervention videos did your child most enjoy? if so, please indicate which. Please select all that apply.

O Instrument Parade

O Song singing

O Actions

O Music Vocabulary

O Rhythm Family

O Rhythm Family Clocks

O Enjoyed all of the above

O Did not enjoy any of the above

Q 3 Please elaborate on your answer.

_____

_____

_____

Q 4 The online class was an important part of the intervention (0 being not at all important and 10 being extremely important).

Not at all Important Extremely Important

0 1 2 3 4 5 6 7 8 9 10

Q 5 Briefly explain your answer.

_____

_____

_____

Q 6 In your opinion, what would be the optimal number of engagements with the video?

O None at all

O Once a week

O Twice a week

O Three times a week

O Four times a week

O Five times a week

O Six times a week

O Every day

Q 7 In your opinion, what would be the optimal length of the music videos?

O 0 minutes

O 5 minutes

O 10 minutes

O 15 minutes

O 20 minutes

Q 8 Any other comments about the online classes and weekly videos

_____

_____

Q 9 Did your child sing or hum any of the songs outside of music class? (0 being that you did not sing the songs at all outside of the class, with 10 being you sang all of the songs outside of the class)

Did Not Sing the Songs Outside Class Sang All of the Songs Outside Class

0 1 2 3 4 5 6 7 8 9 10

Q 10 Please elaborate on your answer.

_____

_____

_____

Q 11 Do you or another guardian use any of these songs at home with your child? (0 being that you do not sing any at all, with 10 being you sing them all of the time)

Did Not Sing Any Sing All of the Time

0 1 2 3 4 5 6 7 8 9 10

Q 12 Please elaborate on your answer.

_____

_____

_____

Q 13 Did you notice any developmental changes during the music intervention? (0 being that you did not notice any changes, with 10 being that you noticed a lot of developmental changes)

Did Not Notice Any Changes Noticed A lot of Changes

0 1 2 3 4 5 6 7 8 9 10

Q 14 If you did, in what areas did you notice the change?

O Communications skills

O Cognitive concepts

O Personal/Social/Emotional capacities

O Motor Skill

Q 15 Please tell us more about any developmental observations in your child.

_____

_____

_____

Q 16 Please tell us anything else that you think is relevant about your child's response to the music intervention

_____

_____

_____

Thank You, You Have now Completed the Questionnaire



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Investigation in the Neurological effect of music on children with Down Syndrome
Daily Diary

Please Enter your Subject ID _____

Please select the date

	Month	Day	Year
Please Select: (1)	▼ January (1 ... December (12)	▼ 1 (1 ... 31 (31)	▼ 1900 (1 ... 2049)

Q1
Engaged with Music Video Today

O Five Minutes - Good Work!

O Fifteen Minutes - Fantastic!

O Ten Minutes Work - Very Good

O Didn't Happen Today Oops, Sorry!

Q2
Sequential Song

O Both singing and actions.

O Observed

O Made no Attempt.

O Singing Only

O Actions Only

Q3
Instrument Recognition

O Attempted but Struggled

O Recognised All

O Attempted with Some Success

O Made no Attempt.

Q4
Instrument Parade?

O Attempted but Struggled

O Attempted with Some Success

O Made no Attempt.

O Recognised All

Q5
Music Note Recognition

O Attempted but Struggled

O Recognised All

O Attempted with Some Success

O Made no Attempt.

Appendix C: Quantitative Analysis and Content Analysis Coding Samples

Why Choose Music?
Coding Sample

Question	Answer	Codes
Reasons for Engaging with Music	• She enjoys it. • He loves music, and it is a positive motivation. • Confidence - to have something they are good at and enjoy doing. Because it's so much easier at that age, and you can always come back to. • Interact with his peer group in a safe setting because he enjoys it • We are singing and dancing at home but have not participated in a music program yet. • We look out for opportunities that are geared towards children like our child to expose them to a maximum of opportunities and experiences. • I hadn't thought if it is benefiting her in another way at the time. • At the start, we thought it would be a positive experience.	Enjoyment Confidence Opportunity Motivation Positive
Reasons for not Engaging with Music	• My child is 4 and has DS. • I feel they are too young and would take up an instrument quicker when she is older. She is involved in a lot of activities at present, including a dance class where she learns the songs she dances to. • COVID • No, they just attend other activities, and I'm conscious of not overfilling his weekends.	COVID Age Other activities Teaching Experience/classes

	• We tried a group session, but my child was unwilling. • They were 3 at the beginning of Covid. Otherwise, we may have enrolled her in a class. • Covid has added to uncertainty around signing up for activities generally. • Concerned that the tutor would not be experienced in teaching a child with an ID. • As a musician, I didn't want to put pressure on them to do music. I wanted it to come from them rather than me telling them that they should. • My child would not be able to keep up with regular music classes.	
Please share any other thoughts or statements regarding the above statements.	• My child has not tried an instrument, as I feel they would pick it up quicker when she is a little bit older. They are already doing lots of activities at present. • He does really enjoy music and dance. • Repetition is extremely important for children with ID. Often, tutors want to move on too quickly, which leads to frustration. • My child really wants to learn to play the keyboard, and it's on the agenda. • Extra time is needed for our kids to get going with a new activity. • Although I cannot play an instrument, I do enjoy listening to music. I think it has huge benefits for children and adults alike. I enjoy singing and dancing with the children in the house and car for fun. I believe that music can release good feelings, promote happy feelings, and encourage self-expression.	Age/older Activities ID Teaching Repetition Enjoy Self-Expression Teaching ID COVID Enjoy

	• Times and locations of music classes are not available, and none are specifically for children with DS, so there is little opportunity to learn. • I tried to involve my child in a music class when he was 6, but they didn't want to join; I hope to try again soon. • The intention is there for both kids to start piano this year. • I attended Music4Children for a few sessions, but the schedule didn't really suit our family.	
Open-Ended Questions Do you feel that the reasons for involving your other child/children in music programmes were any different from those of the child discussed?	• More focus on activities for older siblings due to more time as a parent. Also, few options due to COVID. • She would have less activities at the weekends than him so it nice for her to have her own too. • Yes, they weren't involved in music by choice, my child with DS loves music, but was unwilling to participate in a group session. She was a bit nervous. • No, they are close in age and generally attend activities together. • No, they are close in age and generally attend activities together. • Yes - other children have no interest in music lessons, although they have been offered various instruments, etc. • The older two boys were told that they should learn an instrument which has worked with one but not the other. • I put pressure on my older two to be involved in music.	Activities Group lessons. Age No interest Sports Activities Singing Percussion Piano Enjoy Music TD programmes easier to access. Music as therapeutic

	• No, he enjoys music too, so that would be my main reason for encouraging their interests. • The music programme was for my child with special needs, not his typical siblings. • Yes, other children are very sporty, and that is all-consuming for them (so they don't do music)—music is his thing! • No. • Yes, our daughter did guitar lessons without hesitation as she loves music. • No: my older children enjoyed their involvement in music programmes and went on to learn to play instruments • I think music will be good for the girls. • No, same reasons. I do feel though that my other children would have had better concentration to take part in music programmes • Yes. Our daughter recently started singing lessons as she has a lovely voice. We were considering percussion instruments for our son. • No, I treat both girls equally. • My younger daughter is more eager to get involved in a music programme. • They enjoy music; my eldest plays the piano and plays the ukulele, has a DJ kit, and loves to listen to music. • My other child was younger and is only now turning 6, so he is a good age to learn the piano, although my first child started lessons at 5. • For typically developing children, access to music	

	programmes is much easier, locally. • No • Older children enjoy music in school. • My eldest found playing the piano therapeutic.	
How important do you think music is in a child‘s early experiences?	• COVID did not allow indoor activities such as lessons or music programs. We sang and danced at home until indoor activities allowed us to have a music teacher. • Music is important for speech, mood, concentration, and memory. • Music is extremely important for speech, memory, concentration, and mood. • I feel music is very important as it contributes to speech (nursery rhymes, speech sounds), and it helps concentration and memory. It cheers a child up; they feel like dancing while listening to music. • Children respond and move differently to different music and they enjoy experiencing music. • I found the music very beneficial, and they really enjoy having music as part of their everyday experience. • Very importantly I have always enjoyed music and would like my child to learn and be interested in music. • Of huge value and would love it to be a part of my child's life. • I can appreciate its value and would love it to be a part of my child's life. Despite poor speech, my child is quiet, musical, and sings in tune.	COVID Sing and Dance Speech Mood Concentration Memory Enjoy Value Important Early Years Positive Happiness Well-being Early intervention Self-Expression

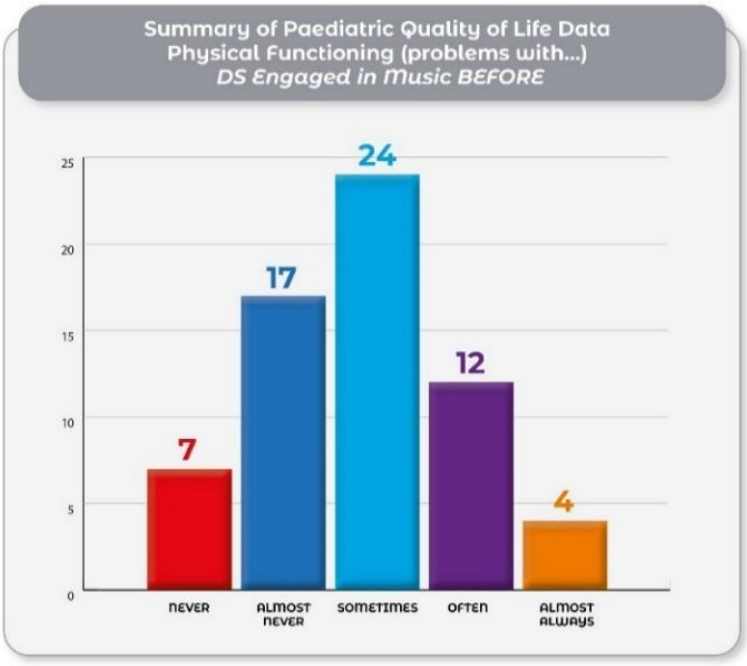
	• I believe it is very important. We have music in the house all the time, and the children love music. We do not play instruments, which has probably led to the children not playing any to date. • I think it is very important, and we have music on in the house a lot. The children love music, but neither I nor the children's father play instruments, which has probably contributed to us not pushing this as much. • As a parent, I am becoming increasingly aware of how important of the role of music in early development • Music is a form of communication. It elicits positive feelings. It stimulates. He likes it. • Benefits personal well-being and general happiness. • It is hugely important. • I feel it is imperative to their development in several ways. However, there are a number of ways in which music can be integrated into a child's life. • It was useful for early intervention to respond to her movements before she spoke to encourage interaction and communication. • Music enhances the children's senses and can incorporate so many early childhood learning skills. • Very important • Music is communication, amongst other things, and aids in development but is also enjoyable and positive. • The child responded to music from a very early age and could	

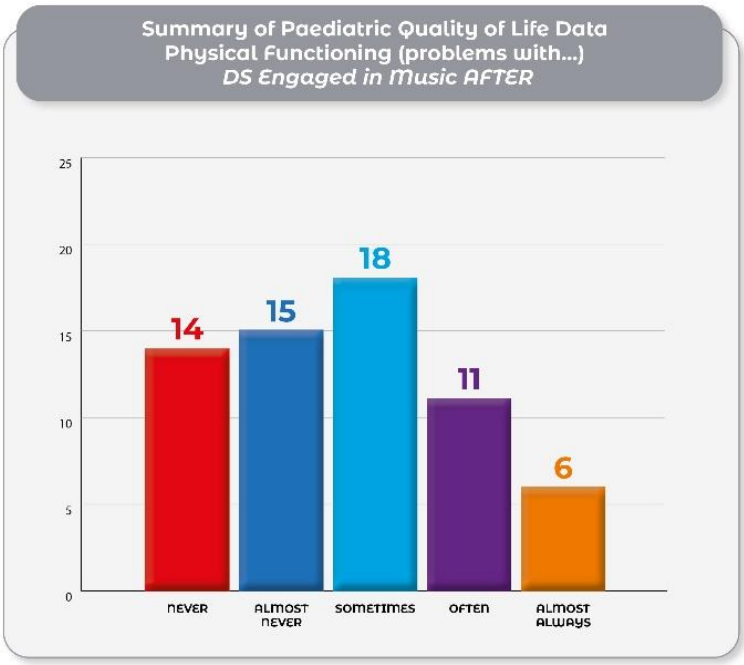
	learn language in conjunction with music. • Music contributes to personal well-being, so it is very important. • Very important as another way of communicating. • All children should have the opportunity to be exposed to as wide a range of music as possible. I would like my kids to enjoy music without feeling limited to any genres. • Very important. • I think it promotes self-expression, confidence, language development and participation in different aspects of family/school life. • We have seen the benefits of music and singing for both of our children, including enjoyment, language learning, speech, diction, memory, and winding down for bed. • Music is a language in itself and aids communication, helps self-regulation and mood and enhances creativity and learning. • I think music opens the soul and creates great pleasure for everyone. • Very important, I believe it's one of the best things to involve them in from an early stage. • Very important. • I think it's very important to introduce all forms of music from an early age. It's fun to sing and dance, and I used music as a way to decompress with my daughter when she was very young. Before she learned how to regulate her big feelings, I would turn on some music and dance with her if I felt she was	

	angry or frustrated and couldn't find the words...or didn't know how to say them. I think music is a great way to express feelings. It resonates emotionally, and some expression through movement in music works. It is great to throw on some tunes while doing the housework...while exercising... I love singing along in the car! • I believe it is important. I see how much happier they are watching/listening to music. • It gives confidence, helps literacy, speech, and social opportunities, and is fun. • Very important, we can see that music helps speech, memory, and recall. • Helps their development. • Music stimulates the brain and makes learning fun. It also encourages children to listen.	
Please share any other thoughts or comments	• Good for involvement and self-growth. • I hope that music will encourage our son's confidence and engagement and show the family that he can engage with them in this form. • Music has featured fully in my child's life. It has helped him stay relaxed and calm, but equally, it has provided great fun and entertainment. Music has helped my child find common ground with peers through dance and expression. • Music was a big part of my life and my sisters when we were children and I appreciate that we were given such an opportunity, I am very keen to provide my kids with the same.	Self-growth Confidence Engagement Calm/Relaxed Interactive Different Teaching Methodologies for Teaching Children with ID Frustration Lack of emphasis Memory

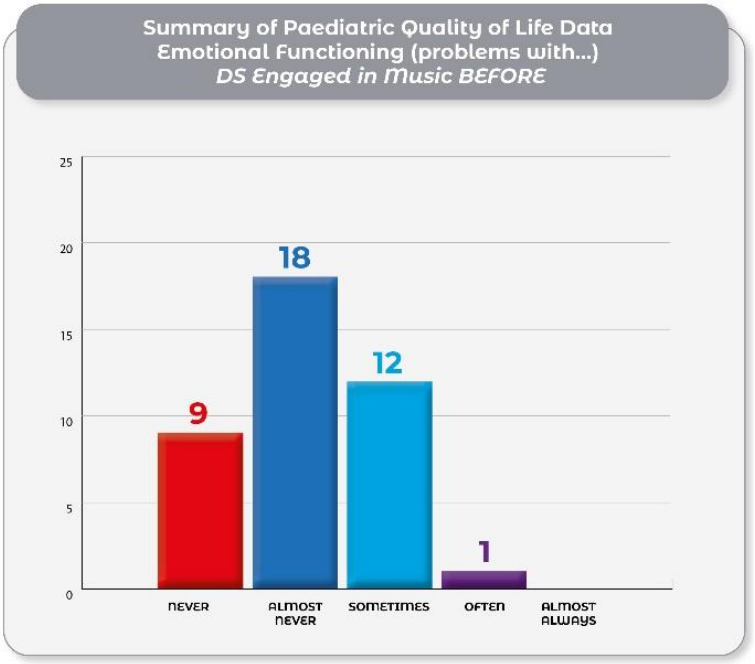
	• I find music very interactive. It's next to impossible not to engage on some level in some way. • Loves music and listens to their favourite songs daily. • Different teaching methodologies are important for children with ID, and many tutors are unaware of this, leading to frustration on all sides! • The lack of emphasis on the importance of music in some schools has undermined its importance in the lives of children as they see it. • As a mum in NICU, I sang one specific song to my child so he would recognise me when I was there.	

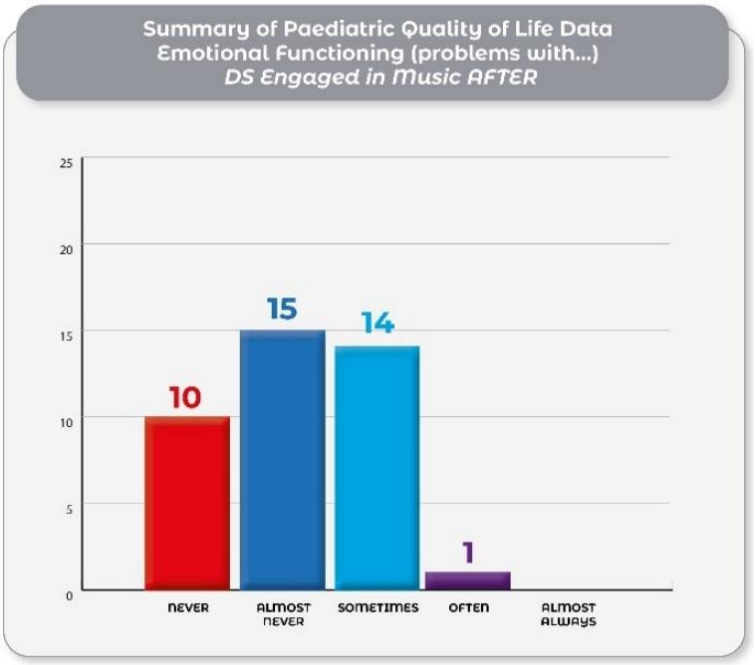
Paediatric Quality of Life: Results



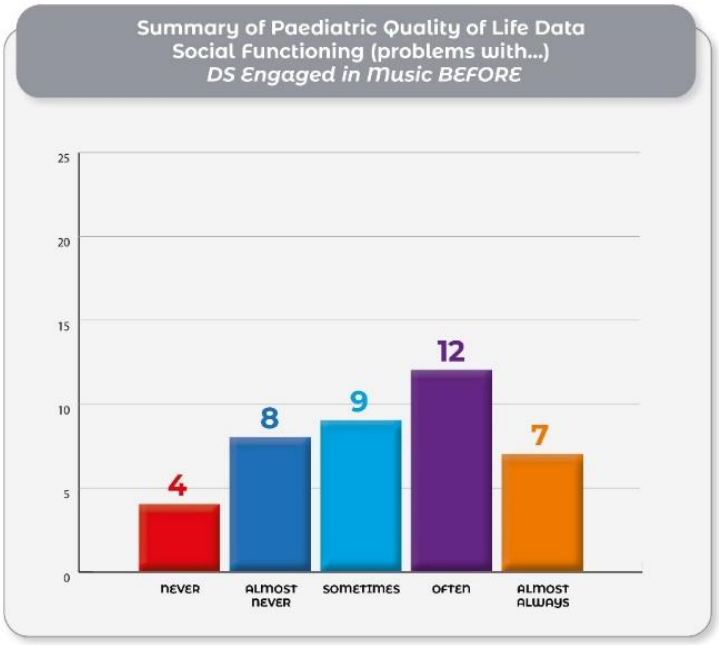


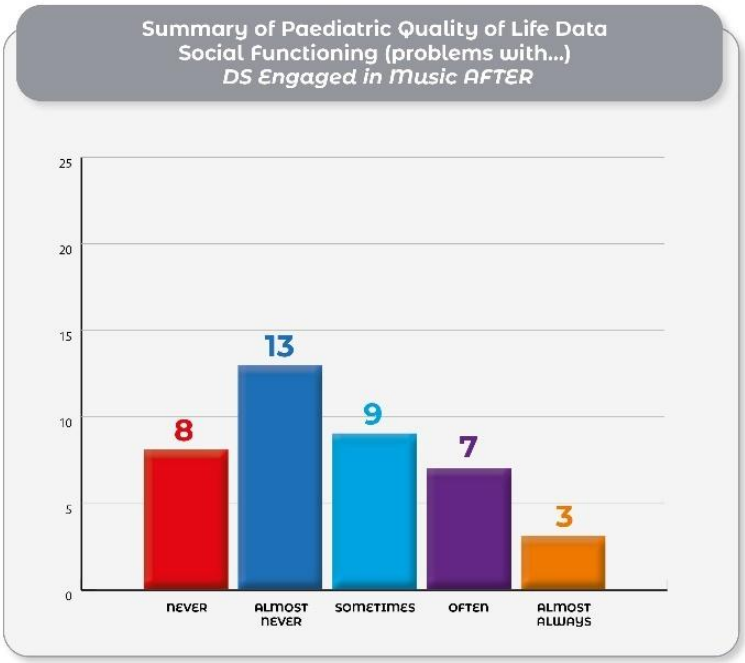
The results of Physical (problems with…) from Children with Down Syndrome engaging with music **before and after** the intervention.



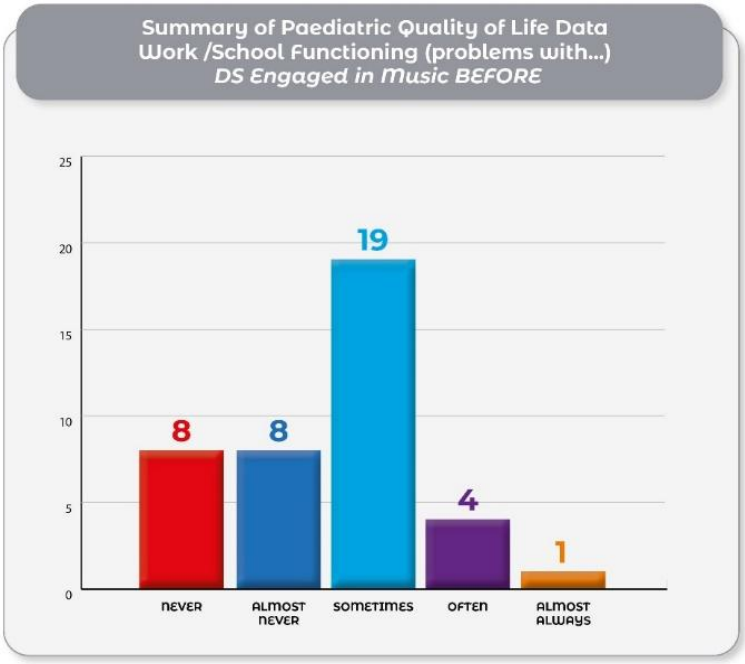


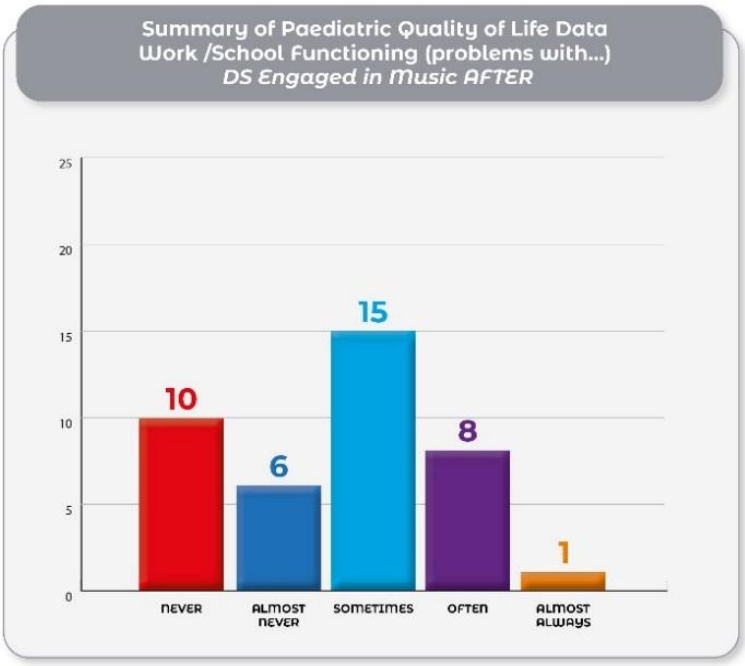
The results of Emotional Functioning (problems with…) from Children with Down Syndrome **engaging** with music **before and after** the intervention.



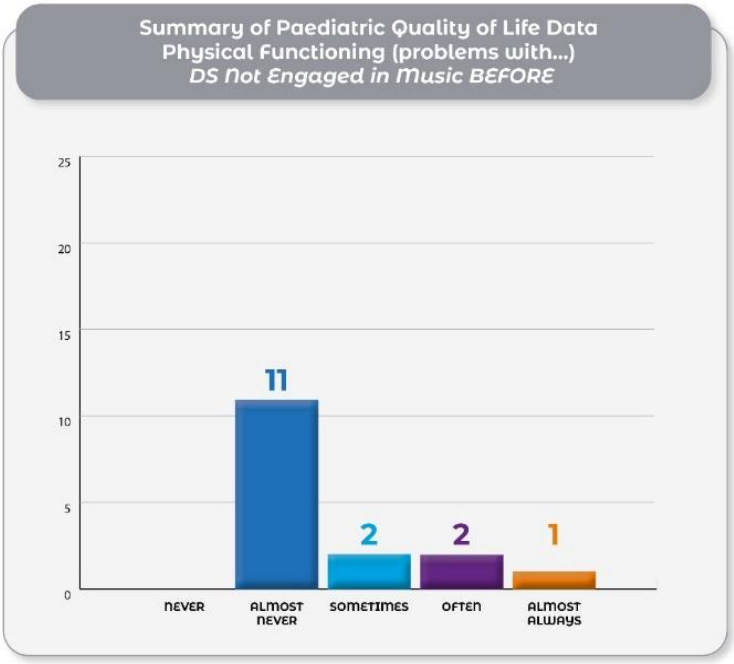


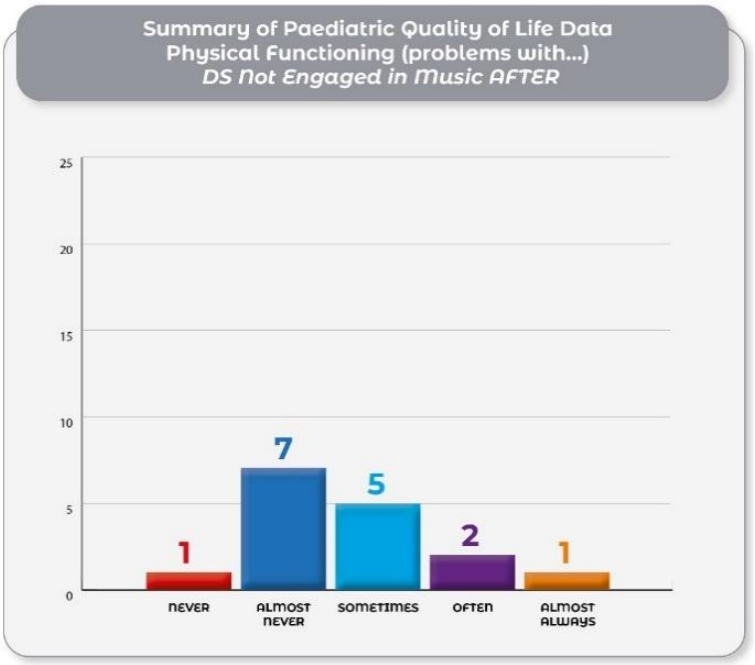
The results of Social Functioning (problems with…) from Children with Down Syndrome **engaging** with music **before and after** the intervention.



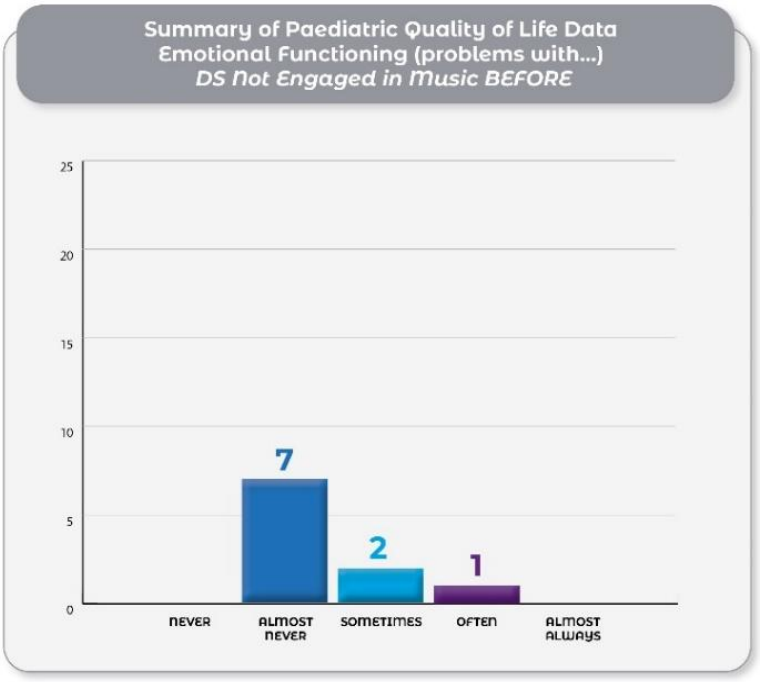


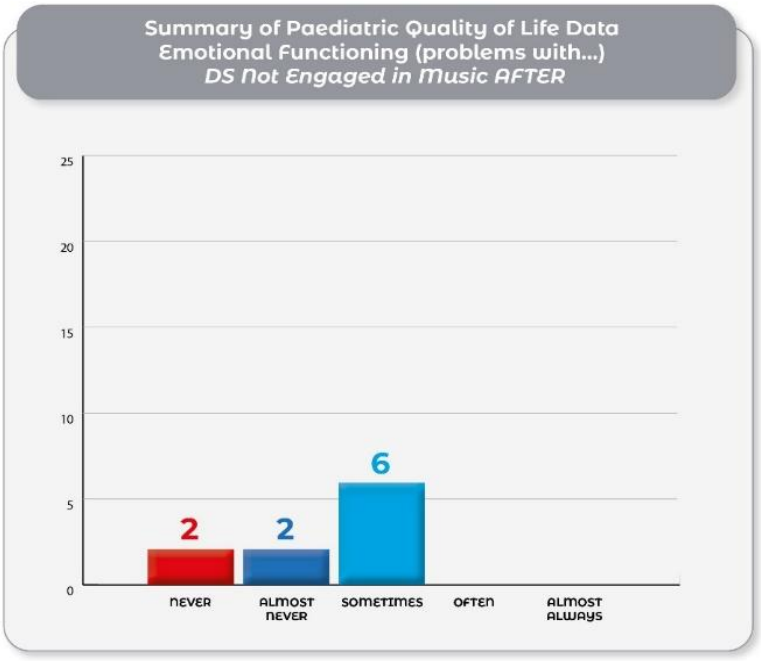
The results of Work/School Functioning (problems with...) from Children with Down Syndrome **engaging** with music **before and after** the intervention.



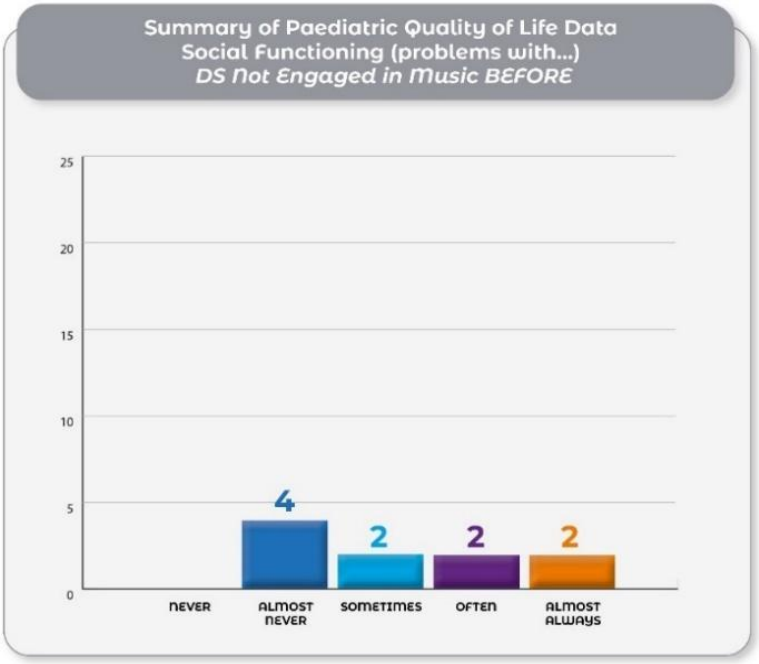


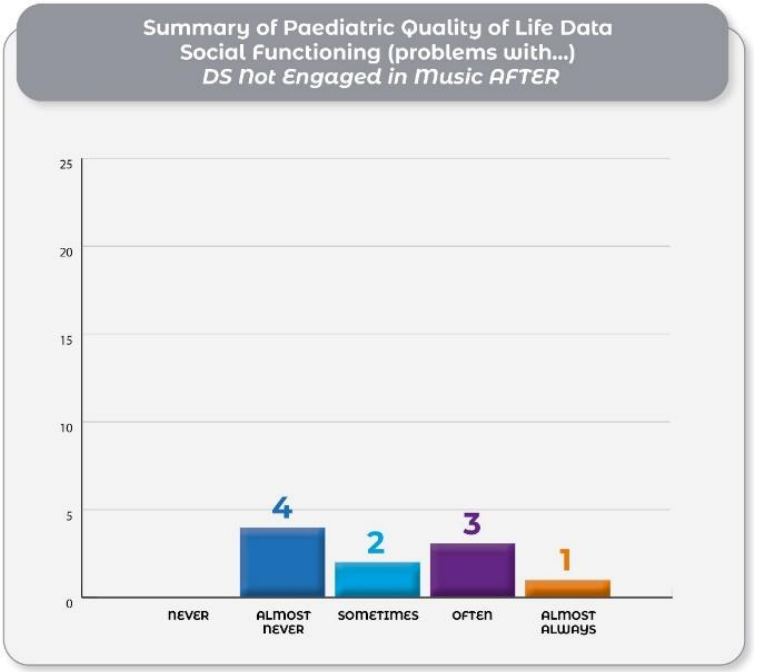
The results of Physical (problems with…) from Children with Down Syndrome not engaging with music **before and after** the intervention.



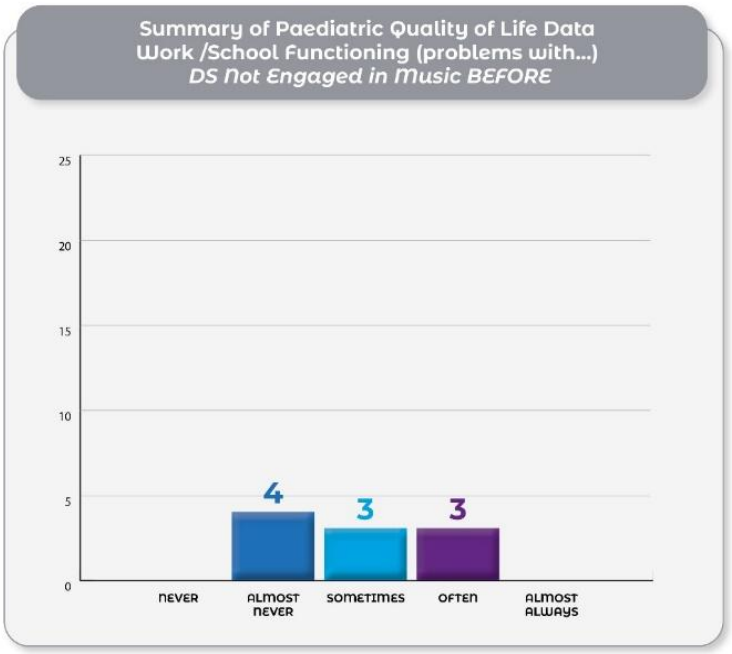


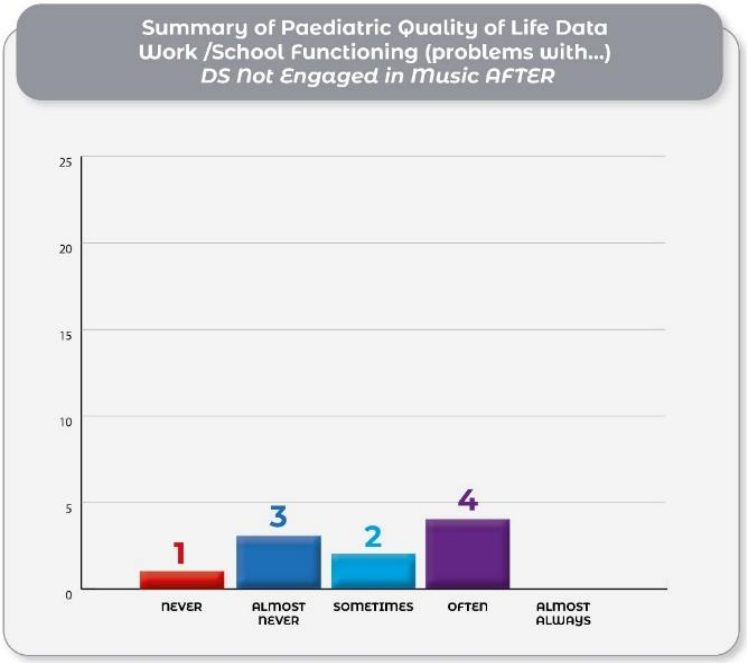
The results of Emotional Functioning (problems with…) from Children with Down Syndrome **not engaging** with music **before** and after the intervention.



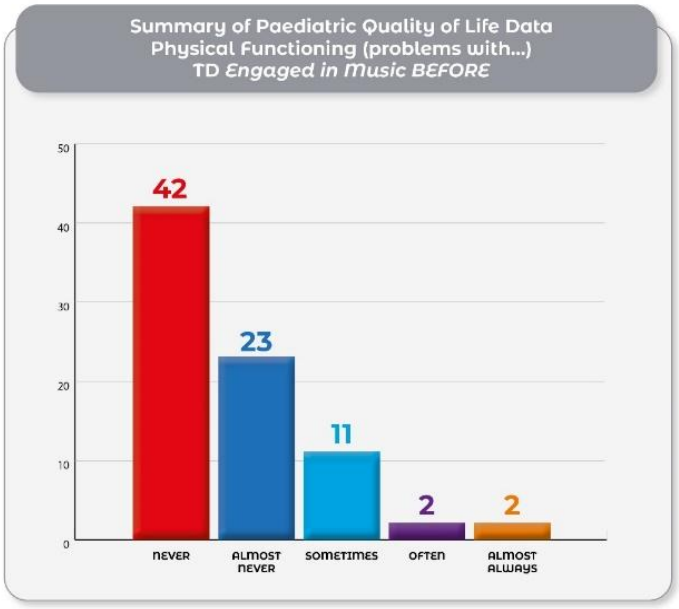


The results of Social Functioning (problems with…) from Children with Down Syndrome **not engaging** with music **before and after** the intervention.



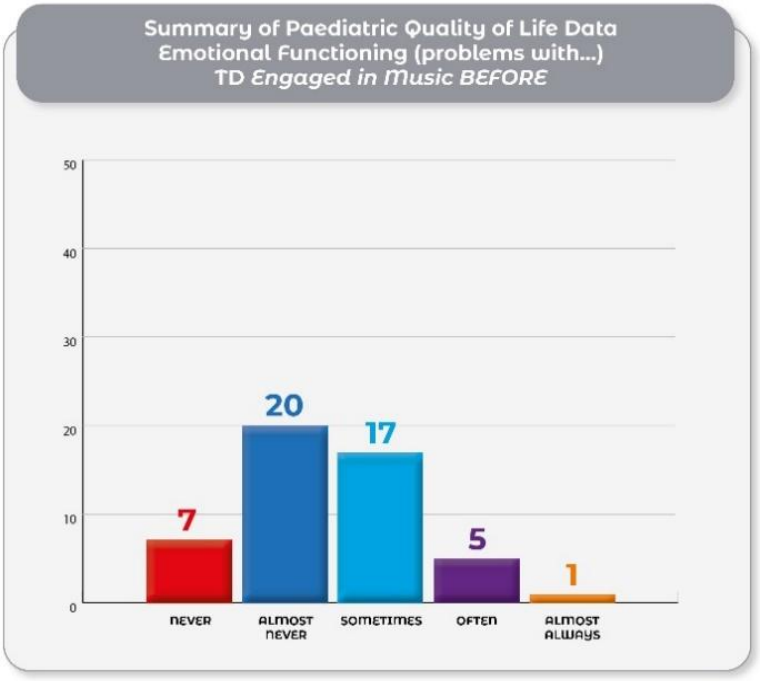


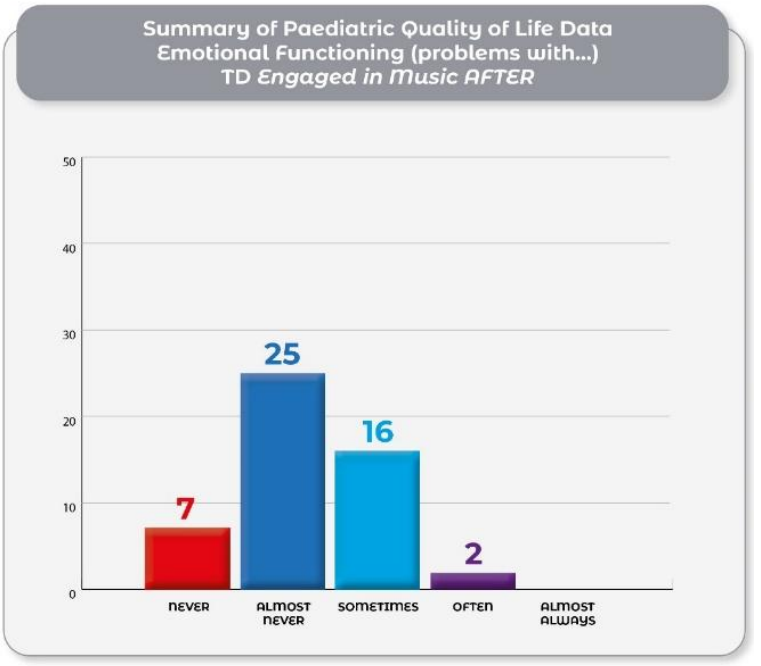
The results of Work/School Functioning (problems with...) from Children with Down Syndrome not **engaging** with music **before and after** the intervention.



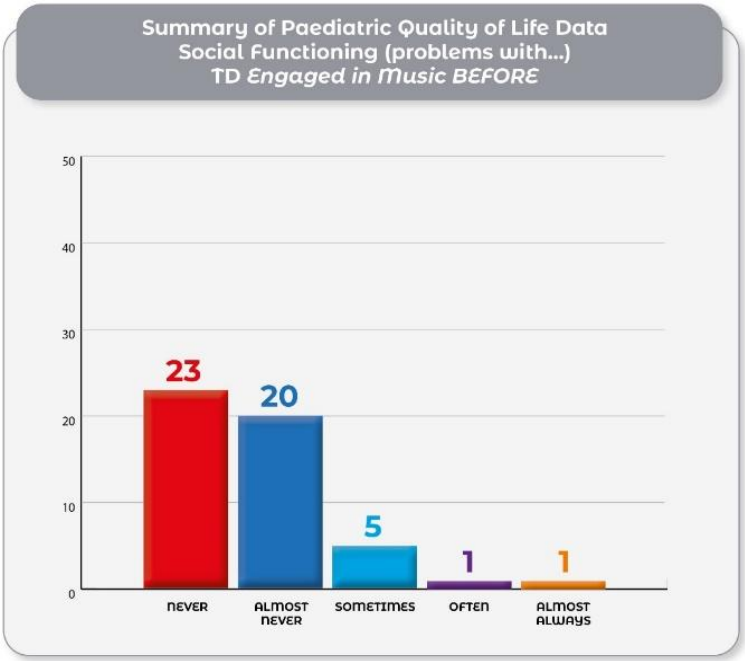


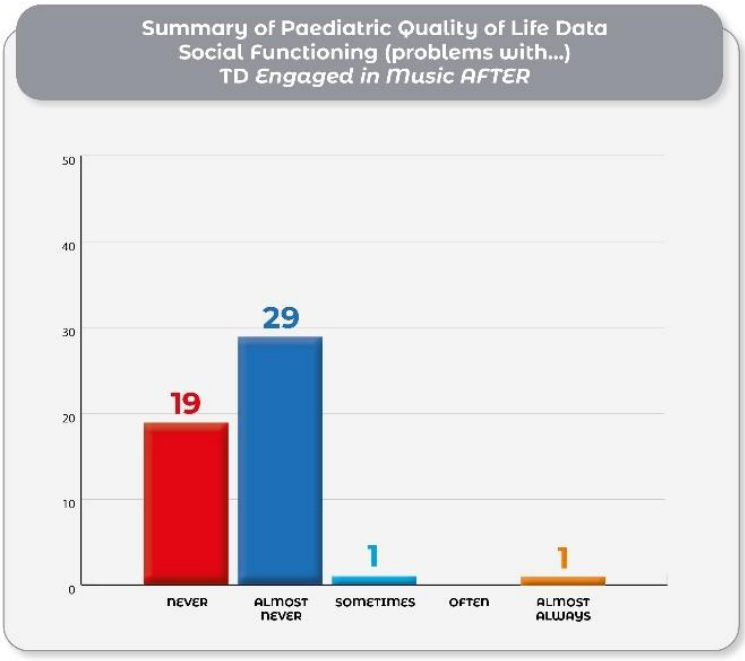
The results of Physical (problems with…) from Typically Developing Children **engaging** with music before and after the intervention.



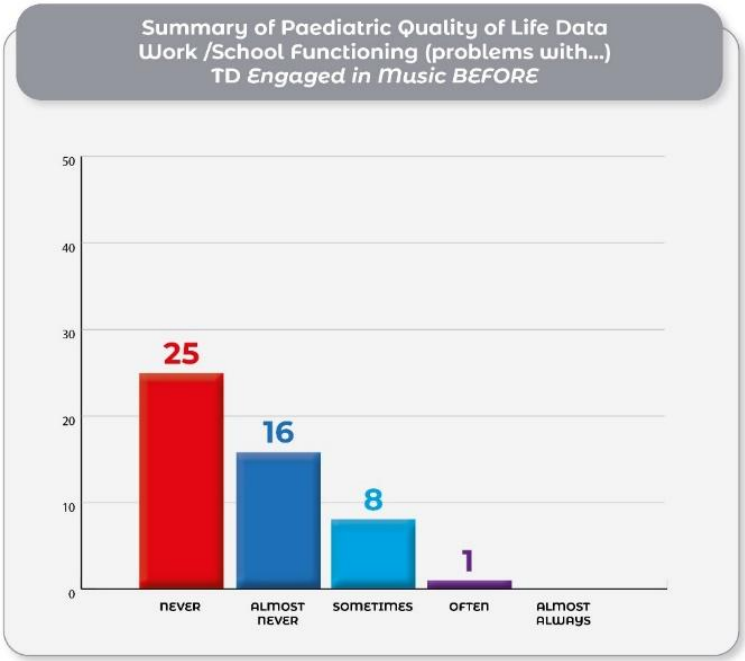


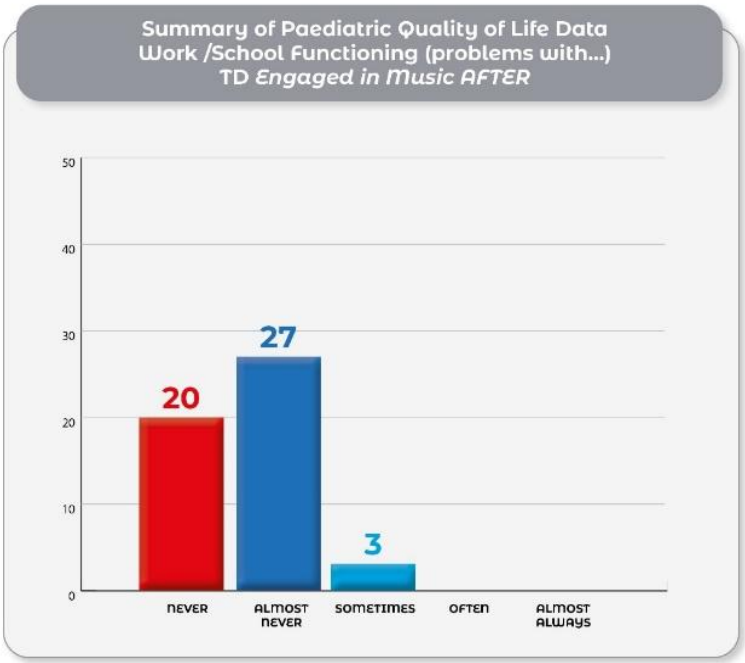
The results of Emotional Functioning (problems with…) from Typically Developing Children **engaging** with music **before** the intervention.



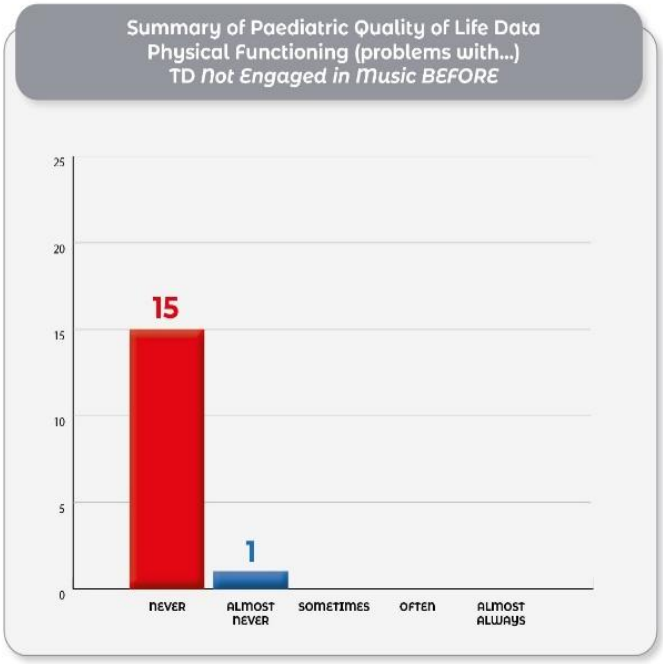


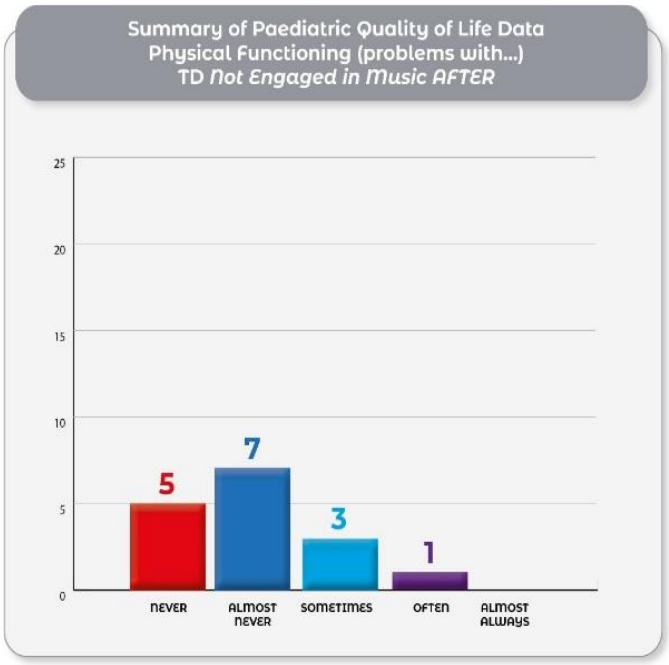
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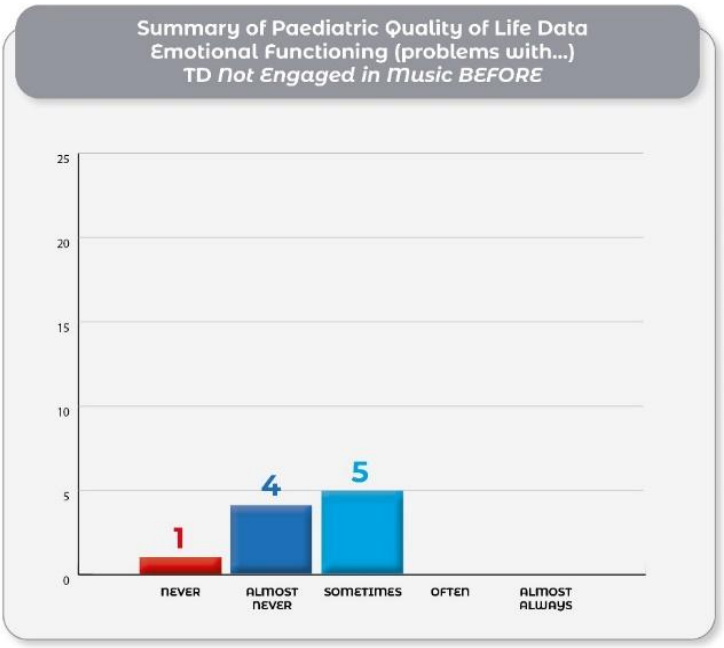


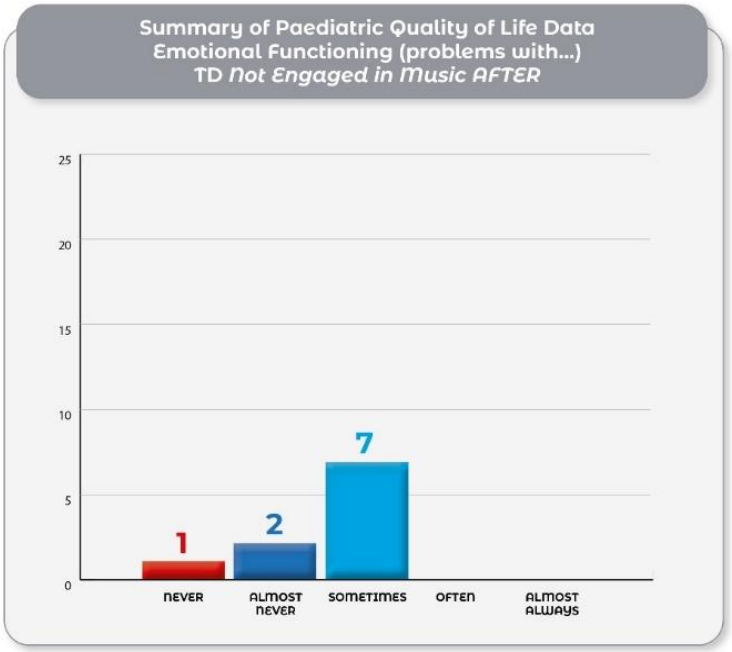
The results of Work/School Functioning (problems with...) from Typically Developing Children **engaging** with music **before and after** the intervention.



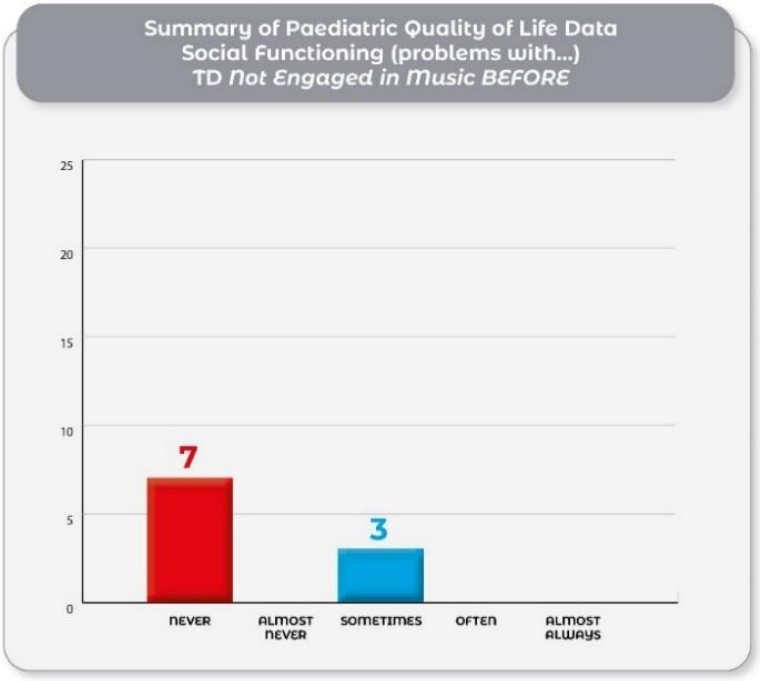


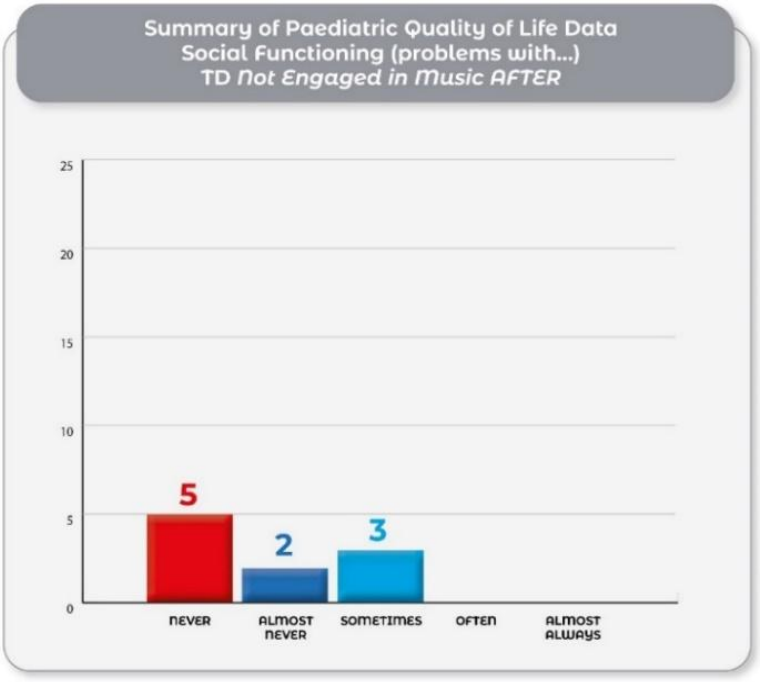
The results of Physical (problems with…) from Typically Developing Children **not engaging** with music **before** the intervention.



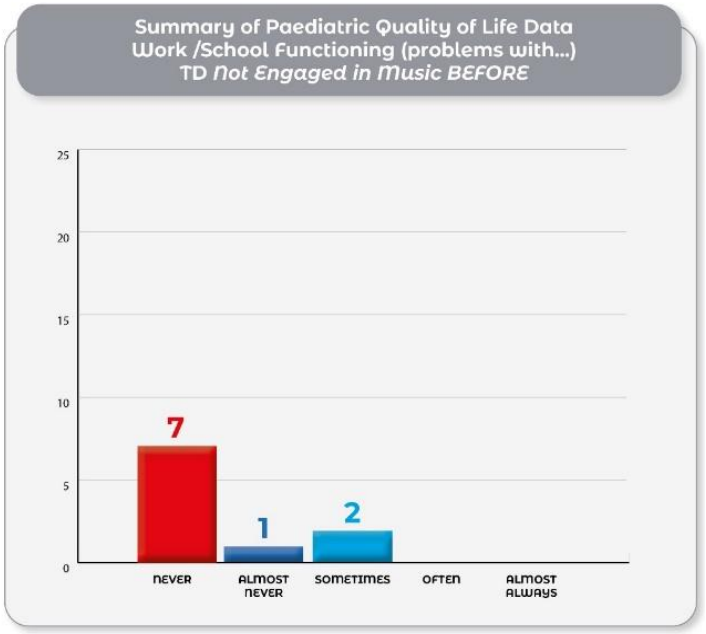


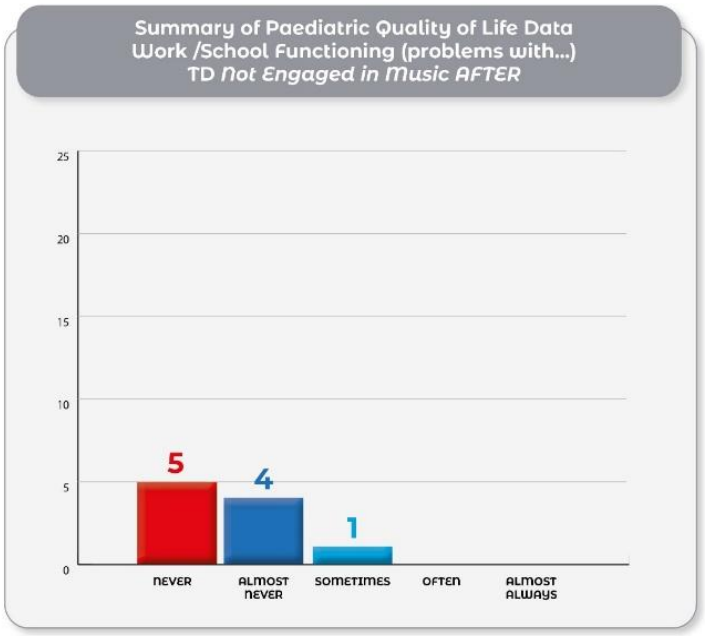
The results of Emotional Functioning (problems with…) from Typically Developing Children **not engaging** with music **before** and after the intervention.





The results of Social Functioning (problems with…) from Typically Developing Children **not engaging** with music **before and after** the intervention.





The results of Work/School Functioning (problems with...) from Typically Developing Children **not engaging** with music **before** the intervention.

Qualitative Description of Your Child's Experience of the Music Intervention: Coding Example

Questions 9 and 10.

Did your child sing or hum any of the songs outside of music class? (0 being that you did not sing the songs at all outside of the class, with 10 being you sang all of the song outside of the class). Please Elaborate on your answer.

Participants' Response	**Codes**
Yes, she was singing the songs to her younger sister. I also heard her using the melodies and putting different lyrics to the songs. She engaged with the songs more so than the instruments.	Singing with Siblings Sociability of Singing Learning through song The adaptability of Song
Very rarely, if ever. Singing isn't something he does often anyway.	Inaccessibility of song
Enjoyed using the instruments to perform.	Enjoyment
My kids still sing and laugh about the songs together and correct each other if one gets a bit wrong.	Sociability of Song - togetherness
Watched them on YouTube himself, also.	Visual Engagement with Song Medium of Intervention
Some songs more than others!	Certain Songs
My daughter is mostly nonverbal, but she would recognise the songs and look to play them on YouTube. Or like when we sung them to her.	Singing as accessibility Medium of Intervention
We all sang a lot of the songs, and the instruments were used lots in day-to-day life (as part of SLT, too!).	Song as Speech Development
The 'High Low' song made an impact particularly! My son would start singing this if the words 'high' or 'low' were mentioned in a sentence!	The transfer of music to language
She liked the songs, and then when family became familiar with them.	Connections through song
Sung them on a Saturday after attending the Zoom lesson.	
I used to hear them singing the songs all of the time, and still do now and again.	Song singing
Some songs were sung by her/him or started by me as it was in my head!	Connections through song

Questions 11 and 12.

Do you or another guardian use any of these songs at home with your child? (O being that you do not sing any at all, with 10 being you sing them all of the time). Please elaborate on your answer.

Participants Responses	**Codes**
Yes, the songs became a part of the household, particularly for the duration of the intervention. We don't sing them as much as we used to now, but sometimes you would hear them being sung.	Family connection through song
Even though he didn't sing them, his younger sibling, who is 3, really loved them. We sang them all the time during the intervention.	Sibling influences
An odd occasion.	Non-transfer
It didn't occur to me!	
Didn't think to!	
We still sing them in the car.	The mobility of song
Those we remembered.	
We like the high-low songs and the forte/piano ones.	Music language
The kids sang them a lot, and it rubbed off on us, eventually.	Family connections through song
We still go back and replay the music intervention videos on a Saturday morning, when their sister is at her music lesson/on a rainy weekend/sick day from school.	Music intervention
Hello everyone song in particular.	Memory and songs
He plays them on Alexa.	
I keep meaning to but forget.	Non- Transfer

Question 15
If you did, in what areas did you notice the change?

Participants Responses	**Codes**
My daughter used a lot of the musical language that she would not have been familiar with before the intervention. So, words like piano/forte and crescendo. Interestingly, these were used only in a musical context and not outside.	Musical Language – frequency infrequency of use.

As the intervention progressed, I felt my son's concentration levels improved slightly. He also used the musical language and interestingly, outside of a musical environment. For example, if someone was being loud, he would say, 'Please don't be forte'. I felt that he engaged with his older sister, who was doing the intervention in a different way. I would hear them talk about what had gone on in the video and ask about the Rhythm family; he would also make up some stories about them during the week.	Integration of Musical Language Piano/Forte and High/Low – the use of Musical Language. Using Narrative as Learning Communication between Siblings
Wanted to show others the instruments.	
I didn't note any changes.	No changed
My son is outgoing and social, but I did feel he gained confidence learning a new language like music and that confidence made him more vocal. He wanted to share the learning and show off what he knew. He also had loads of chats with his sister about what they were learning and the two of them could be heard singing and dancing to the songs they learned along the way.	The Benefits of Learning a Musical Language Shared learning Communication between siblings Song as a sociable interaction
Attempted to say the music vocabulary.	The Impact of Musical Language
The main thing would be concentration, recognising the videos, and knowing when they were over.	Music and heightened concentration
Overall calmer and more open to instruction.	Music and Learning
Communication skills (language): my son learned and retained the instrument names and music terms (e.g. crescendo, largo, presto).	The Positive Impact of Music on Speech Development
Motor skills (gross motor): My son kept time to the music perfectly (shaking, clapping, stomping). Motor skills (fine motor): My son learned to use the space bar to stop the video (to find an instrument/ pipe cleaner (music note)) and restart. He has progressed to using the computer mouse!	Music and Motor Skill Development
First time using a computer for learning. First time hearing musical terms.	The Impact of Online Learning

Developed a greater understanding about music, vocabulary associated with music, notes and how music is made.	The Positive Impact of Music on Vocabulary.
I just noticed they loved the routine of the video every night but didn't like the online class, probably because they're not used to Zoom.	The Impact of Online Learning
It's hard to tell what a natural progress was or due to the intervention, but she made strides in her schoolwork, communication skills, listening and learning.	Music on Learning, Communication skills

Question 16

Please tell us anything else that you think are relevant about your child's response to the music intervention.

Participants Responses	**Codes**
The enjoyment factor was really great to see. Also, how much can be learned? She made strides in her schoolwork, communication skills, listening and learning through the videos more than in the online class. Seeing the contrast between these two forms of delivery was interesting. While digital technology and devices are criticised a lot, it was great to see how they can also positively impact learning. Rosie really engaged with the presenters on the screen. She was less engaged with the body percussion exercises. But overall, really loved the programme.	The Impact of Online Learning – Positive versus Negative Formats of Delivery The Role of music in learning. Screen play Musical mediums of engagement
Relaxed her more.	The Role of Music on Relaxation
This child was 8 at the time of the study and I would say some of the content was a bit juvenile for her. She enjoyed it overall but wasn't as engaged as my 5-year-old. My son just loved it. Outside of other commitments and activities playing into the schedule, he really enjoyed the music, and he now will sit at the piano (which arrived at XMAS) and has the confidence to bang out a made-up tune. He wouldn't have done that before. It's definitely given him some musicality. He enjoyed the experience, still watches some	The Impact of Music Content on Engagement Music and Confidence Music and Instruments

of the YouTube clips and plays with the egg shaker and tambourine.	
When I would say 'Music Class', she would go and get the music bag, which was a great association for her.	Music and Memory
My son loves music. He sings and dances all the time - at home, in the car, in school. We were surprised and very impressed at his ability to remember the names of the people in the rhythm family, the tempo, the instruments, the music terms. Thanks so much to Eva for giving us this opportunity. We are on the lookout for bodhrán lessons locally as a result	The Impact of Music on Learning Music and Memory. Music Interventions and Learning
Carryover of the learning was noticed with later comments from the child using the words piano and forte.	The Impact of Music on Language Development
They found Padraig very funny and loved when he acted silly in the videos.	The Role of Play in Learning. Musical Play Laughter in Learning.

Appendix D: Narrative for Cognitive Tests

Narrative Created and Used for Temporal Order Judgement Task and Spatial Memory Test

Phase One

Hello, boys and girls. I wonder if you can help me.
My little friend Mert the mouse has run off, and I don't know where they are.

I wonder if you can help me. Mert is out in the garden, and they love playing tricks, so it is really hard to find them. However, if we use our listening ears, we might be able to find the mouse, as sometimes they make a squeak coming from different parts of the garden.

Can you help me by **pressing the arrow on the right when you hear Mert on your right side? When you hear Mert on your left side, press the left arrow**.
Thank you for your help!

Press the Space Bar, and you will be ready to go!

TAKE A BREAK

PHASE TWO

Well done on trying to find Mert, but this mouse is super quick, and I think we will need more help as it is now getting very dark outside. I was wondering if we could use a flashlight. We might be able to find Mert easier. You will see your light above or below a crossroads near my house where the mouse might have escaped.

Be sure to let me know where they might be. **Press the left arrow when the light flashes above the crossroads and press the right arrow when the light flashes below**.
Good luck. I hope we find Mert.

Press the Space Bar, and you will be ready to go!

TAKE A BREAK

PHASE THREE

Thanks so much for helping us find Mert, but the mouse is still hiding from us—they are such tricksters!!! I think we will now have to use both our flashlight and our listening ears to find Mert.

When you hear Mert squeaking or see your flashlight first at the crossroads, **press the left arrow if you hear him squeak** and press the **right arrow if you see the flash.**

Press the Space Bar, and you will be ready to go!

TAKE A BREAK

PHASE FOUR

Well done!
Thank you so much, everyone. You have helped me find Mert. After Mert's afternoon of excitement, poor Mert is very hungry. I wonder what Mert would like to eat. Hmm, do you think they would like apples? What about burgers and chips? No, you are right! Mert Loves Cheese!!!

Mert is very particular about how they like to eat the blocks of cheese. The blocks must be in the right order. Can you help Mert to put these cheese blocks in order?

Press the Space Bar, and you will be ready to go!

THANK YOU

Appendix E: Music Intervention Details

Sound Bag Contents

Information Sheet Streamers Lollipop Sticks Pipe Cleaners Kazoo Tambourine Egg Shaker Rhythm Family Portrait Circle Activity Sheet Balloons A4 Paper

Lesson One

Hello Song Introduction to the Instruments using the **clock** to show the children when to play and when not to play. **(Clock 1)** Introduction to the **Rhythm Family** by clapping and using **Dalcroze-**inspired movements. Now play the **Rhythm family** on the tambourine and egg shaker Musical extracts are played on the piano using rhythmic values – which rhythm is it? Let's make **Granny Semibreve** out of a pipe cleaner. Short **Instrument Parade** (introducing sequence recognition) – tambourine, egg shaker, kazoo. Zippidy Zap Song. Goodbye Song

Lesson Two

Hello Song Revise the **Rhythm Family** by using body percussion and Instruments. Use the clock that has the **Rhythm Family (Clock 3)** Play the following pieces of music **Haydn's Surprise Symphony, Toreador's Song from Bizet's Carmen, Canon in D by Pachelbel**) and invite the children to guess which music belongs to which Rhythm Family Member by using streamers. Let's Make **Cara Crotchet** out of a pipe cleaner

Introduce a short **Instrument Parade** (introducing sequence recognition) – tambourine, egg shaker, kazoo, sleigh bells.
Zippidy Zap Song
Goodbye Song

Lesson Three

Hello Song
Choose your favourite instrument and revise the rhythm in the clock **(Clock 3)**
The **Rhythm Family** is going on holiday. Introduce Mrs Treble's House and project the musical stave on the board behind it.
Introduce the High Low Song
Repeat the High Low Song game using the streamers. Move the streamers up high for the high notes and down low for the low notes.
Let's Make **Mr Minim** this week.
Introduce a short **Instrument Parade** (introducing sequence recognition) – tambourine, sleigh bells, triangle, guiro.
There's A Beat in My Feat Song (using Kazoos)
Goodbye Song

Lesson Four

Hello Song
Introduce more **Body Percussion** by singing Are You Sleeping?
Play A Choir Making Sounds of the Rain
Using blank the A4 paper 'Let's Go for a walk' – inspired by **Dalcroze.**
Introduce the terms ***Piano/Forte***
Play the ***Piano/Forte Song***
Let's Make the **Quavers** out of pipe cleaners this week.
Introduce a short **Instrument Parade (**introducing sequence recognition) – tambourine, egg shaker, sleigh bells, kazoo, agogo, triangle. After the children repeat the sequence, take one instrument away. Let them guess which instrument is missing.
Let's make the **Quavers** out of pipe cleaners.
There's A Beat in My Feat (using Kazoos)
Goodbye Song

Lesson Five

New Hello Song is introduced
Play instruments with the **Clock 2**
Revise ***Piano*** and ***Forte*** Song
Ask the children what Instruments play ***Forte*** and which ones play ***Piano***. The ***Forte*** instruments include the timpani, flute, trumpet, and trombone, while the ***Piano*** instrument is the flute.
Explore ***Piano*** and ***Forte*** in the orchestral work entitled William Tell by Rossini.
It is holiday time again, and the **Rhythm Family** are going to **Mrs Treble's House.**
In preparation for the visit to **Mrs. Treble's House,** revise the **Rhythm Family** on **Clock 4**.
Introduce C, D and E – (Cora, Dora, Eva). What song has three notes?
Introduce a short **Instrument Parade (**introducing sequence recognition)– tambourine, egg shaker, sleigh bells, kazoo, agogo, triangle. After the children repeat the sequence, take two instruments away. Let them guess which instruments are missing.
Music Music Song (Lollipop Sticks)
Goodbye Song

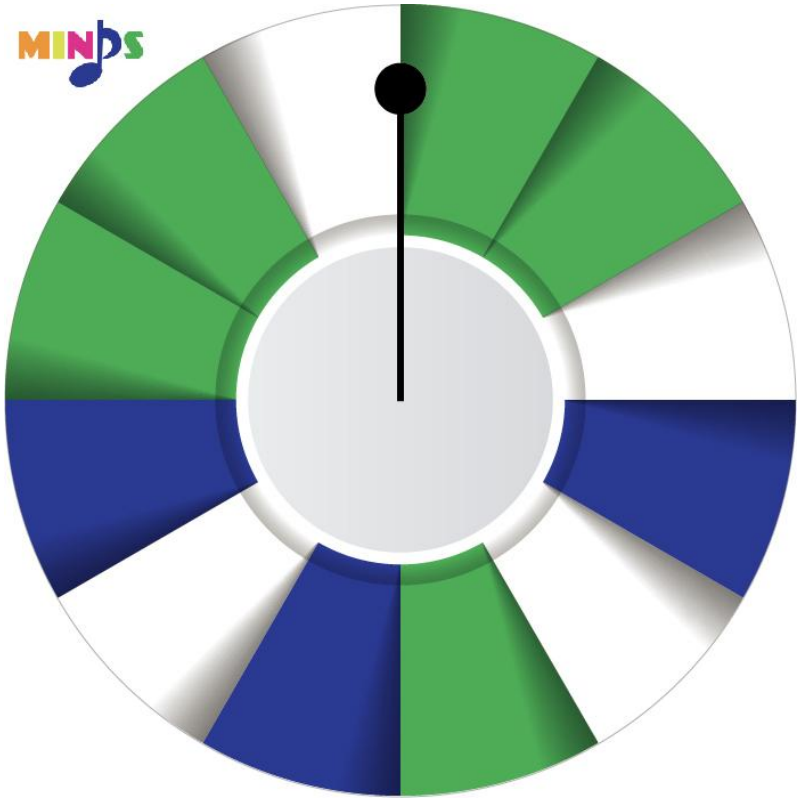
Lesson 6

The New Hello Song
Revise the **Rhythm Family** using **Clock 4**
Refer to the big **C*rescendo*** in the ***Forte*** song and look at the big **C*rescendo*** on the board behind.
Introduce the ***Crescendo* Song**.
Play Grieg's In the Hall of the Mountain King to explore the C***rescendo*** in the third movement into the fourth, keeping the beat (do you remember there was a beat in our feet?)—this piece of music gets louder.
The **Rhythm family** is still on holiday.
Cora, Dora, Eva, Fia, Gina are in the house (Notes C, D, E, F, G), but Dora and Fia had to go out. What does the piece of music sound like now? Sing Twinkle Twinkle Little Star
Introduce a short **Instrument Parade (**introducing sequence recognition)– tambourine, egg shaker, sleigh bells, kazoo, agogo, triangle. After the children repeat the sequence, take three instruments away. Let them guess which instruments are missing.
Music Music (Lollipop Sticks)
Goodbye Song

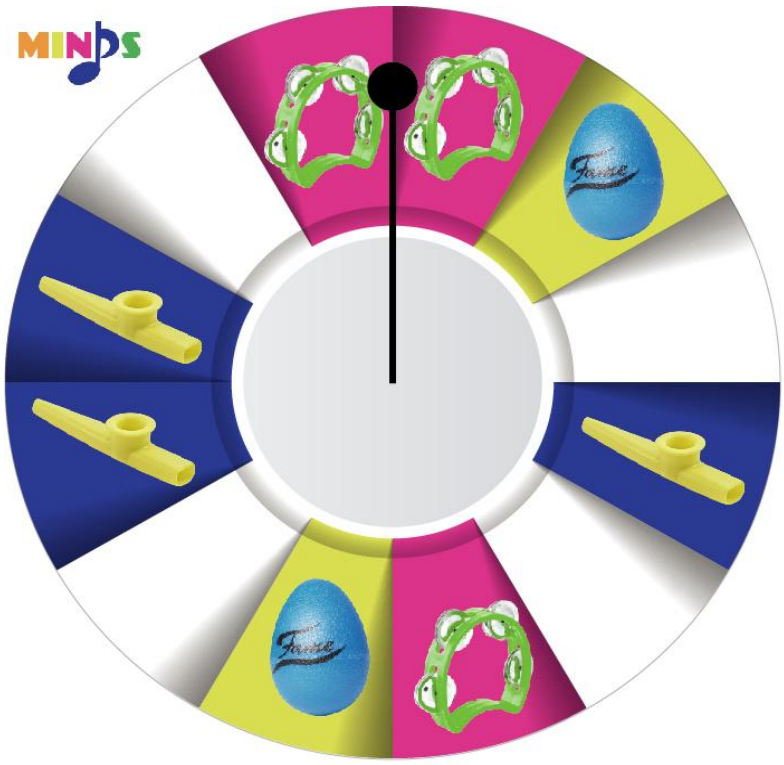
Lesson Seven

The New Hello Song
Do as I'm Doing Song, alternating between Fast or Slow ***Tempi***
Introduce the ***Presto*/L*argo*** song
Can you guess which music is ***Presto*** and ***Largo***? Play the Mexican Hat song (Jarabe Taptio).
Introduce **Musical Form** by using the picture of a Sandwich
Play the Let's Make a Sandwich song
Revisit the Mexican Hat song and discuss **A and B Form**
Play the Twinkle Twinkle Little Star Game
Revise the **Rhythm Family** by using **Clock 4**
Revise: **Mrs Trebles House**
Introduce a short **Instrument Parade (**introducing sequence recognition)– tambourine, egg shaker, sleigh bells, kazoo, agogo, triangle. After the children repeat the sequence, take three instruments away. Let them guess which instruments are missing.
Stand-Up Sit-Down Song
Goodbye Song

Rhythm Family Clocks



Clock One: Different sounds are played on the green and blue colours; everyone must be silent when the clock hand lands on the white © Music4Children.



Clock Two: Egg Shakers are played when the clock hand reaches yellow, kazoos when it reaches blue, and tambourines when it reaches pink.



Clock Three: When the hand lands on a member of the rhythm family, the participant must play the relevant rhythmic value.



Clock Four: This is the most advanced version of the clock that contains many more rhythms and the clock time moves faster.

The Rhythm Family



GRANNY
SEMIBREVE

MINDS

The Rhythm Family



MR. MINIM

MINDS

The Rhythm Family



CARA
CROTCHET



MINDS

The Rhythm Family



KATIE AND
CAOIMHE
QUAVER



MINDS