Enhancing Communication in Girls with Rett Syndrome through Songs in Music Therapy

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ENHANCING COMMUNICATION IN GIRLS WITH RETT SYNDROME THROUGH SONGS IN MUSIC THERAPY

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ABSTRACT

Rett syndrome is a neurological disorder resulting from an X-linked dominant mutation affecting mainly females and found in a variety of racial and ethnic groups worldwide. One of the main areas of affected functioning for females with Rett syndrome is a severe impairment of receptive and expressive communication, and as a result educational intervention in this area is considered to be of utmost importance. Females with Rett syndrome have been observed as very responsive to music and, used therapeutically by trained practitioners, this intervention has been shown to promote and motivate their desire to interact and communicate with their surroundings. In order to investigate the use of music therapy as a means of promoting communication, seven girls with Rett syndrome, ages 4-10 participated in a study over eight months. A single case, multiple probe design was used, where during 30-minute trials, three times per week involved the choice of and observed responses to familiar and unfamiliar songs.

Results show that children with Rett syndrome have intentional choice making and an ability to learn and sustain learning over time. Musical analysis and observed responses revealed clear preferences for certain songs and non-preference for others, consistent between all seven participants, where preferred songs have certain identifiable musical features. All the participants in the present study revealed a variety of consistent behaviors and emotional responses demonstrating intentional communication.
I would like to acknowledge and thank Prof. Tony Wigram at Aalborg University who has followed, supervised, inspired and supported me and the research project from the beginning till the finish product. His endless hours of dedication, patience and humor, his wise remarks and broad knowledge kept me always thinking and broadening as this project unfolded.

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I dedicate this work to these girls and their families.
INTRODUCTION

Rett syndrome is a neurological disorder resulting from an X-linked dominant mutation (Amir et al., 2000), affecting mainly females and found in a variety of racial and ethnic groups worldwide (Hagberg, Aicardi, Dias, & Ramos, 1983). It is a frequent cause of neurological dysfunction in females, accounted for the most common cause for multiple-disability among females (Hagberg, 1985b).

Individuals with Rett syndrome have special educational needs, and they experience the most profound and extreme learning disability (Lewis & Wilson, 1996). Due to the fact that one of the main features of this population is a severe impairment of receptive and expressive communication (typically with no expressive use of verbal language), communication has been targeted and considered as a priority within the main educational intervention (Sigafos & Woodyatt, 1996). Traditionally, the primary focus of communication intervention for the developmentally disabled was to enhance speech. Throughout the years, many contributors expanded the scope of communication and language intervention from primary focus on the expressive aspects of communication (learning to talk) to a broader emphasis on the multiple process of communication, including receptive communication (expanding comprehension) (Iacono, Carter, & Hook, 1998; Siegel-Causey & Bashinski, 1997).

From reviewing current literature, it is clear that in addition to lacking basic communication abilities as a result of their primary limitations, individuals with severe communication impairment are handicapped by the lack of sufficient opportunities for indicating preferences and making choices. It is believed that the experience in choice making has not been provided to them by their surroundings (Bambara, Koger, Katzer, & Davenport, 1995; Sigafos, Laurie, and Pennell, 1995). "Potential Communicative Act" suggested by Sigafos et al., (1999, 2000) acknowledges the possibility that existing informal and idiosyncratic behaviors could become effective forms of communication.
Research and information about the cognitive and communicative potential of females with Rett syndrome is scarce and sometimes show contradicting and indefinite results (Sigafoos et al., 1999; Van Acker, 1996; Woodyatt & Ozanne, 1992, 1994).

It has been indicated that people with Rett syndrome communicate by using non-symbolic means such as gestures, vocalizations and body positioning (Coleman, Brubaker, Hunter, & Smith, 1988), and by retaining positive contacts to people's faces/eyes reacting with smiles (Burford & Trevarthen, 1997; Sigafoos et al., 1999).

Females with Rett syndrome have been observed as very responsive to music. When music is used therapeutically by trained practitioners, this intervention has been shown to promote and motivate their desire to interact and communicate with their surroundings, as well as to develop their cognitive, affective, sensory-motor and physical skills (Allan, 1991; Coleman, 1987; Elefant & Lotan, 1998; Hadsell & Coleman, 1988; Hill, 1997; Kerr, 1992; Lindberg, 1991; Merker, Bergstrom Isacsson, Witt Engerstrom, 2001; Montague, 1988, Takehisa & Takehisa Silvestri, n.d.; Wesecky, 1986; Wigram, 1991, 1995; Wigram & Cass, 1996).


I have been working with girls with Rett syndrome for the past 13 years. During this period the hidden skills of this population in learning and communicating as well as their profound attraction to music became clear to me. Clinical experience has shown me that once children with Rett syndrome find an interesting and motivating environment, such as music therapy, they are likely to become very
involved, attuned and more likely to learn. It is therefore natural that music should be utilized as a medium for promoting communication for individuals with Rett syndrome. This notion was clarified when I began working intensively at a center for developmentally disabled children “Beit Issie Shapiro” in Raanana, Israel where seven girls with Rett syndrome were enrolled as students, and as a result provided the foundation for this study. While many publications anecdotally reported the value of music therapy for this population, there was also a significant paucity of clinically controlled trials demonstrating efficacy, and sustaining of effect over time.

The purpose of this research was therefore to investigate the following question: Can songs in music therapy enhance communication in girls with Rett syndrome? The sub questions supporting the primary research question were:
1. Are girls with Rett syndrome able to make intentional choices?
2. Are girls with Rett syndrome able to learn, and sustain learning over time?
3. Do girls with Rett syndrome reveal consistent preferences through choices they make?
4. How do girls with Rett syndrome demonstrate emotional and communicative behaviors?

An overview of the contents of this thesis describes the presentation of information in a format typical of experimental studies and includes the following:

Chapter 1 documents a literature review which, examines past and current knowledge on Rett syndrome, communication, music therapy and the interaction between these three areas, leading up to the above mentioned research questions.

Chapter 2 gives a detailed description of the specific research method and design. The stages of the experimental trials are described (baseline, probe, intervention, and maintenance) during which the responses of each participant were recorded for analysis and interpretation.
Chapter 3 presents the results and outcomes of the intervention with seven girls with Rett syndrome. Data gathered from intentional choice making, response time, learning process, song preference, and emotional, communicative and pathological behaviors would be presented, analyzed and interpreted.

Chapter 4 presents the results and discussion of song analysis. An in-depth analysis of the musical elements of the songs used in the present study was undertaken to elicit key elements that had an influence on the research population, and can be used in futuristic clinical and research settings. This analysis was not proposed as part of the study, but has been included as a post hoc analysis together with a discussion to further explain the likely basis behind the consistently preferred and non-preferred choices. Song performance as it was presented in this study will be discussed at the end of this chapter.

Chapter 5 discusses the findings using the same framework as Chapter 3. Any weaknesses and limitations of the research design are considered, and clinical application together with the need for further studies is discussed in relation to intentional choice making, response time, learning process, emotional, communicative and pathological behaviors.

The thesis will end with a short summary of the main research findings, and offers conclusion to the study.
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CHAPTER ONE

1.1 RETT SYNDROME

INTRODUCTION

This study was concerned with the effect of music therapy on Rett syndrome. As this syndrome is sometimes less well known among some professionals, it is necessary to begin with a short introduction of Rett syndrome - pathology, characteristics, and current knowledge - in order to provide the reader with an up-to-date review of the subject. My review will begin at the discovery of Rett syndrome.

Rett syndrome is a neurological disorder affecting mainly females and found in a variety of racial and ethnic groups worldwide. Rett syndrome was first described by Dr. Andreas Rett (Rett, 1966). It received worldwide recognition following a paper by Dr. Bengt Hagberg and colleagues (Hagberg, Aicardi, Dias, & Ramos, 1983) and the first International Congress was held in 1985 (Hagberg, 1993).

Childhood development in females with Rett syndrome proceeds in an apparently normal fashion during pregnancy and over the first 6-18 months, at which point development comes to a halt, they regress and lose many, if not most, of their acquired skills (Graham, 1995). The research and information about the cognitive and communicative potential of females with Rett syndrome is sparse and sometimes reports are contradicting and with indefinite results. There seems to be an agreement that although prior to the regression the girls appear to develop according to the developmental milestones (Amir et al., 2000), a closer look reveals small abnormalities that can be detected by experienced health professionals (Kerr, 2001).
Thereafter, there is a rapid deterioration and loss of acquired speech and purposeful hand use. Individuals with Rett syndrome exhibit one or more stereotyped hand movements such as hand wringing, hand-washing, hand-clapping, and hand-to-mouth movements (Ishikawa et al., 1978; Weiss, 1996). The repetitive hand movements appear involuntary and occur during most of their periods of wakefulness.

The deceleration of head growth, and jerky body movements of the trunk and limbs accompany the developmental deterioration in individuals with Rett syndrome. Typically, they present with a broad base gait and swaying movements of the shoulders when walking (Kerr & Stephenson, 1986). Many girls begin walking within the normal age-range for walking, while others show significant delays. Some begin walking and may lose this skill, while others will continue walking throughout life (IRSA, 1997).

Other physical problems, such as seizures, scoliosis, and breathing abnormalities may appear. The seizures can range from non-existent to severe, but seem to lessen in their intensity in later adolescence. The scoliosis is a prominent feature in females with Rett syndrome but may vary from mild to severe. Breathing abnormalities may occur, but can also decrease with age. These may include: hyperventilation, breath holding, apnea and air swallowing, which may result in abdominal bloating (IRSA, 1997, 2000). Apraxia (dyspraxia), the inability to program the body to perform motor movements, is the most fundamental and severely handicapping aspect of Rett syndrome. It can interfere with all body movement, including eye gaze and speech, making it difficult for individuals with Rett syndrome to execute what she wants to do.

Rett syndrome has been most often misdiagnosed as autism, cerebral palsy, and non-specific developmental delay or as a result of the care of overly apprehensive mothers (Leonard, Bower, & English, 1997). However, increasing knowledge and
the ability to determine pathology by blood analysis in a majority of cases has improved diagnostic accuracy.

While some health professionals may not be familiar with Rett syndrome, it is a frequent cause of neurological dysfunction in females, accounting for the most common cause for multiple-disability among females (Hagberg, 1985b), or the second most common cause for severe mental retardation in females after Down’s syndrome (Ellaway & Christodoulou, 2001).

The prevalence among different countries ranges from 1:10,000 - 15,000 (Hagberg, 1985b; Kerr & Stephenson, 1985) to 1: 22,800 females (Kozinetz et al., 1993). Most researchers now agree that Rett syndrome is a developmental disorder rather than a progressive, degenerative disorder as once thought. Despite illness and other possible complications, survival into adulthood is usually expected. Although the genetic source for Rett syndrome has been discovered, it is not possible to detect the affected gene in all females with Rett syndrome. Due to this fact, the Rett syndrome diagnosis relies heavily on diagnostic criteria developed by Dr. Hagberg and Dr. Witt Engerstrom of Sweden (Hagberg, Goutieres, Hanefeld, Rett, & Wilson, 1985). These criteria have been mildly changed over the years by the International Rett Syndrome Association's (IRSA) professional advisory board consisting of well-known experts in the field of Rett syndrome (IRSA, 1997).

1.1.1 Diagnostic Criteria

The official definition of Rett syndrome in DSM IV (1994) is presented below (in italics). The expanded and full definition of the disorder is written according to the International Rett Syndrome Association (IRSA), since the organization closely and constantly updates the diagnostic criteria by world experts on the syndrome.
Definition by the DSM IV:

“Rett’s disorder: A pervasive developmental disorder characterized by the appearance, between 5 and 48 months of age, of decelerated head growth, loss of previously acquired purposeful hand movements and development of stereotyped hand movements (hand wringing), loss of social engagement (social interaction may develop later), poorly coordinated gait or trunk movements, and impaired expressive and receptive language with severe psychomotor retardation. Development during the first 6 months of life appears normal” (DSM IV, 1994).

The following are the diagnosis criteria required for the recognition of Rett syndrome after the exclusion of other handicapping conditions (IRSA, 1997, 2000):

- A period of apparently normal development between 6-18 months.
- Normal head circumference at birth followed by slowing of the rate of head growth with age (3 months - 4 years).
- Severely impaired expressive language and loss of purposeful hand skills makes assessment difficult of receptive language and intelligence.
- Repetitive hand movements including one or more of the following: hand washing, hand wringing, hand clapping, and hand mouthing, which can become almost constant while awake.
- Shakiness of the torso, which may also involve the limbs, particularly when upset or agitated. If able to walk, unsteady, wide-based stiff-legged gait/toe-walking.
1.1.2 Supportive Criteria

The following symptoms are not required for the diagnosis, but may also be seen as secondary symptoms in some girls. These features may not be observed in younger girls, but may evolve with age (IRSA, 1997, 2000):

- Breathing dysfunctions, which include breath holding or apnea, hyperventilation and air swallowing that may result in abdominal bloating and distension.
- EEG abnormalities - slowing of normal electrical patterns, the appearance of epileptiform patterns and loss of normal sleep characteristics.
- Seizures.
- Muscle rigidity/spasticity/joint contractures, which increase with age.
- Scoliosis (curvature of the spine).
- Teeth grinding (bruxism).
- Small feet (in relationship to stature).
- Growth retardation.
- Decreased body fat and muscle mass (but tendency toward obesity in some adults).
- Abnormal sleep patterns and irritability or agitation.
- Chewing and/or swallowing difficulties.
- Poor circulation of the lower extremities, cold and bluish-red feet and legs.
- Decreased mobility with age.
- Constipation.

Not all individuals with Rett syndrome do display all of these symptoms, and individual symptoms may vary in severity.
1.1.3 Stages in Rett syndrome

Rett syndrome appears in four consecutive stages (IRSA, 1997, 2000):

**Stage I**: "Early Onset Stage" appears after a period of apparently normal to near normal development (until 6-18 months). This is the period were the symptoms of Rett syndrome may just emerge and there may be delays in normal development of gross motor milestones. This period is quite short and may last a few weeks to a few months.

**Stage II**: "Rapid Destructive Stage". During this period there is a rapid (or in some cases more gradual) regression in which the child loses her acquired speech and purposeful hand movements. This stage appears between the ages of 1-4 years and may last a few weeks to several months. This period is especially turbulent and the stereotyped hand movements will emerge. Rett syndrome has sometimes been misdiagnosed during this stage due to similarity in some of the pathological characteristics with children with low-functioning autism. The child may appear autistic-like with the loss of social interaction and communication skills. The child may avoid eye contact, avoid auditory input, and lose interest in playing with toys as they lose purposeful hand movement. During this period the child will typically have episodes of screaming and sleeping disturbances. Breathing irregularities may appear with breath holding or hyperventilation. Head circumference will fall on the "percentile chart".

**Stage III**: "Plateau Stage". This stage is long and quite stable stage and can last for many years. Seizures, apraxia and motor problems are more prominent, however the child seems less irritable, more alert and interested in her surroundings. This is the period were the child may be accessible to basic learning skills. The introduction and development of non-verbal communication strategies is most likely to become effective and helpful from this stage on.
Stage IV: "Late Motor Deterioration Stage"
Stage IVA (Previously ambulant)
Stage IVB (Never Ambulant)
This period begins at age ten and is characterized by reduced mobility and the loss of ambulation. However there is no decline in cognitive, communication or hand skills. Repetitive hand movement may decrease and scoliosis is a prominent feature.

The above diagnostic criteria are helpful in detecting the young child with Rett syndrome, however there have been reports of ‘under’ diagnosis Rett syndrome even by trained specialists (Leonard et al., 1997). At a very young age while signs of Rett syndrome are still obscure and inconclusive, detecting the syndrome might not be an easy task. During puberty adolescence and adulthood, diagnosis becomes harder due to a reduction of some of the more obvious and well-recognized pathological characteristics in favor of more generalized global developmental delay.

The discovery of the gene for Rett syndrome in September of 1999 (Amir et al., 2000) provided greater scientific potential for detection and diagnosis for almost everyone involved in this debilitating syndrome.

1.1.4 Genetics in Rett syndrome

There has long been a debate as to whether or not Rett syndrome is a genetic disorder. In September of 1999, the discovery of a genetic mutation (MECP2) on the long arm of the X chromosome (Xq28) revealed significant insight into the cause of Rett syndrome. This mutation has now been found in up to 90% (Amir, 2001) of typical and atypical cases of Rett syndrome. The consensus hypothesis is that Rett syndrome is an X-linked dominant disorder caused by new mutations in
the Rett syndrome gene 99% of the time. There are only a small number of familial Rett syndrome recurrences (1% of the reported cases) and those are mostly sisters or twins. There are some families who have Rett syndrome in two generations (aunt-niece) (Amir et al., 2000).

Since the discovery of the gene, 23 boys with Rett syndrome have been identified internationally (Hoffbuht, 2000; Kerr, 2001). When Rett syndrome occurs in males, it is usually lethal, causing miscarriage, stillbirth or early death (Hoffbuht, 2000). Continued research will focus on other still unidentified genetic factors that contribute to Rett syndrome. Researchers agree that the severity of Rett syndrome is probably not linked to the exact location of individual mutations on MECP2, but to the X inactivation patterns in each affected girl (Amir et al., 2000).

To date there is no known remedy that can repair the neurological damage caused by the genetic fault of Rett syndrome. No medical solutions have succeeded in alleviating symptoms connected to Rett syndrome during pre-, peri- or postnatal periods and no subsequent medical treatment that will improve the physical condition and the functional abilities in this population (apart from the anti-epileptic medication for the ones with seizures). Therefore, different types of remedial therapy are likely to be more important for any potential improvement, and even the slowing of deterioration. Some researchers believe that knowledgeable therapy might change the course of Rett syndrome (Jacobsen, Viken, & von Tetchner, 2001). What can be done to help these children? What is the common treatment approach suitable for individuals with Rett syndrome? What are the main goals of the different therapeutic approaches for this population? The next section will offer possible answers to all of these questions.
1.1.5 Treatment strategies for Rett syndrome

Due to the complexity and the challenging disabilities of Rett syndrome, all known treatment approaches (and some less familiar) have been attempted to aid the needing child and her family. This section will discuss therapeutic and educational approaches for the Rett syndrome population.

1.1.5.1 Therapeutic intervention for individuals with Rett syndrome

Treatment approaches for the individuals with Rett syndrome are intended at elevating the individual’s quality of life through enhancing her abilities, reducing disease, and adding content to the individual, usually seen wondering about the room as if without aim.

Music therapy - Dr. Rett recognized the potential of music therapy to penetrate the heavy shield of the disability masking potentially hidden abilities of the child with Rett syndrome (Rett, 1982). Music therapists use their applied musical skills of improvisation and songs to establish rapport with the child, utilizing it as bridge to bond and engage in non-verbal forms of communication. Music therapy can open channels, which can offer a child the opportunity, through musical sounds, to express emotions and feelings, further enabling some communication and learning to take place. Current knowledge on the use of music therapy as an intervention for the child with Rett syndrome, suggests that music therapy can enhance functional hand use and reduce stereotypic hand movements, develop possibilities for choice making, enhance vocalization, improve attention and eye contact, develop emotional-communicative channels and facilitate in relaxing and soothing the child (Allan, 1991; Coleman, 1987; Elefant & Lotan 1998; Hadsell
Physiotherapy - Due to the fact that girls with Rett syndrome present with a variety of physical impairments, and that the progression of this disorder repeatedly show periods of deterioration, physical therapy is considered an important intervention for the child with Rett syndrome. The few articles suggesting physical therapy management (Beuchel, 2001; Elefant & Lotan, 1998; Hanks, 1990, 1996) emphasize the importance of individual evaluation as the key point of personal adapted intervention. Nevertheless, several similarities between females with Rett syndrome suggest the following points of commonality in many girls receiving physiotherapy: Develop and maintain walking ability, develop and improve transitions, prevent and reduce contractures, reducing agitation, enhancing independence, building, adjusting and helping in purchasing accessories (Lotan, 2001).

Hydrotherapy - Most girls with Rett syndrome will need physical treatment throughout their lives, although movement and external facilitation are usually disliked by most of them. This conflict raises the need for a mediating surrounding that can ease and reduce the fear of movement typical for Rett syndrome. An environment encompassing physical treatment with soothing effects is the warm relaxing water of the hydrotherapy pool. Although there are differences in the appearance of the symptoms of Rett syndrome in each child, possible mutual treatment directions have been repeatedly identified in several girls with Rett syndrome: Improving A.D.L. (Activities of Daily Living), facilitating hand use, preventing deterioration and maintaining orthopedic status, building up breath
control and improving cardio-vascular abilities (Hunter, 1999; Lotan, 2001).

**Occupational therapy** - One the most debilitating aspect of Rett syndrome is apraxia. Handling apraxia is one of the many areas occupational therapy has to offer the child with Rett syndrome. Occupational therapy as part of a team approach enables the child with Rett syndrome more possibilities in assembling her diffused body scheme and organizing it for the day-to-day situations. The occupational therapy basic work is to ‘adapt’ whether it is the activity, materials or the environment (Hunter, 1999). Possible therapeutic options are available for the occupational therapist working with Rett syndrome: Improve A.D.L., organize and improve sensory perception, enhance manual function, estimate and enhance cognitive function.

**Speech therapy** - Most girls with Rett syndrome are unable to speak. Few have the ability for expressive language. The use of sign language seems to have very small chance of achieving positive results due to the poor hand function characteristic of Rett syndrome and due to the apraxia, which makes imitation an impossible task. Taking such condition into account, the speech therapist working with the child with Rett syndrome tends to concentrate on achieving maximal communication abilities through the use of alternative and augmentative communication. A second field where the speech therapist’s expertise can be of use is in organizing the child’s mouth in order reduce eating problems so common in Rett syndrome (Budden, 1995). Possible goals for speech therapy with the Rett population tend to be: Adjusting the adequate communication method for each child, educating staff members and family in maximal use of communication devices enabling maximal
communication level, improving oral functioning for eating and vocalization. It is important to mention the fact that literacy is an expanding area of success for children with Rett syndrome (Hetzroni & Rubin, 1998) and a collaborating team of teachers and speech therapist might enhance the child’s development in this field.

**Hippotherapy** (therapeutic horseback riding) - Girls with Rett syndrome are known for their love of animals (Hunter, 1999; Levinson, 1996). The sensation of touching the warm horse’s body and the normality of riding a horse are entwined into the experience of movement while riding the horse’s back makes this experience a whole and accepted treatment for individuals with Rett syndrome. Possible goals for hippotherapy treatment are the organizing body perception and helping in building body scheme, improving functional hand use, improving balance reaction and social interaction.

Due to the debilitating nature of Rett syndrome and the inability of conventional therapy management to offer successful intervention programs, there have been anecdotal reports on different methods that seemed to yield positive results for the child with Rett syndrome.

- **Domen De Lacato** - Physical intervention management has managed to teach one 18 year old girl with Rett syndrome to walk (Rettnet, 1997).

- **Osteopathic treatment** - Such as Myo-Facial Release and Cranio-Sacral therapy have been reported anecdotally to improve the quality of life for several children with Rett syndrome (Rettnet, 1997).

- **Irlen lenses** - Originally used with children with learning disability and reported anecdotally to improve reaction speed and achieve higher spirits
for two girls with Rett syndrome (K. Johnson, personal communication, March 18, 1997).

- **Lovas therapy** - A behavioral modification management reported as achieving positive results with one child with Rett syndrome (Rettnet, 1997, 1998).

- **Pet therapy** - Several reports have conveyed the power of animals (dogs, dolphins and others) on the mood and cooperation of children with Rett syndrome (Chess, 1997; Levinson, 1996).

- **Aromatherapy** - A single report of aromatherapy with massage had been administered and reported on success in reducing hand stereotypical movement and improving circulation to the feet (Price, 1996).

The above reported intervention approaches are but a drop in a vast ocean of therapeutic interventions applied to this population and are utilized as a result of the limited progress seen when the child is exposed to formal treatment. All of these approaches are meant to achieve progress in areas such as well-being, quality of life, or daily life functioning.

### 1.1.5.2 Educational approaches for Rett syndrome

Individuals with Rett syndrome have special educational needs, and they experience the deepest, most extreme learning disability. The ‘right’ approach to the disabled child cannot be standardized from procedures documented in books and in the literature, it is found in the child’s interactions with her parents, teachers and therapists throughout her daily experiences. The child with Rett syndrome is influenced by (and through) her encounters with different people at home, at school
and in the community, but these people are not acting in a vacuum. They represent the atmosphere around special education and special children.

Three decades ago children with learning disabilities were perceived as uneducable, but these attitudes have rapidly changed over the last two decades (Lewis & Wilson, 1996). It is frequently suggested that children with different developmental disabilities can achieve more than has been anticipated or expected. Not only can they participate in their family’s activities, attend school, enjoy the company of friends, and leisure time activities, they can become an integral and contributing part of their community (Puescel, Bernior, & Weidenman, 1988 in Lewis & Wilson, 1996).

Such change of attitudes towards the children with disabilities as a whole have provoked changes in legislation, requiring that all children with developmental disabilities receive education suited to their level of development. The framework for this research is placed within a center where educational opportunities for girls with Rett syndrome rely on potential for development. To promote communication, and utilize what limited communication skills are there, educators increasingly resort to all potential methods of non-verbal and gestural communication. Therefore, any potential benefits from a study such as this relies on the future application of communication-based strategies that assume comprehension and expression. Teachers frequently have an extended role in educating children with Rett syndrome that includes:

- **Empowering and promoting good relationships with the child’s parents.** If we take into account the fact that most of the child’s days are spent with her family; it should be obvious that educational direction and goals must be shared with all that come in contact with the child, enabling a steady and consistent advancements.
- **Flexibility** with the understanding that each child is different and unique, thus requiring special (individually oriented) education. This need is strongly advocated in the Rett syndrome population.

- **Leading** a team of professionals can bring out the hidden talents of the child with Rett syndrome.

- **Stamina and patience** is necessary since the severe nature of the disorder can sometimes results in one step forward being followed by five steps backwards. Such regression, and eventually severely limited progress, can cause despair in even the most enthusiastic professionals.

- **Maintaining a close and supportive partnership** between educational team members (including parents) can facilitate insightful solutions to a frustrating regression. Educational programs for all children with special needs are planned through joint decision making of all those concerned with the care of the child (parents, teachers, caregivers and therapists).

- **Knowledge of Rett syndrome** will help the educational team in expecting the unpredictable liability of mood and the functioning level typical to the population, and will help in planning appropriate responses to occurring events and future developments.

- **Holding an appropriate evaluation** for the child’s specific needs, will direct the educational and therapeutic team in the specific treatment plan for each child.

Goals for females Rett syndrome are usually within the area of function, social skills and communication (Hunter, 1999; Lewis & Wilson, 1996; Lotan, 2001).
Due to the fact that one of the main features of females with Rett syndrome is the impairment of receptive and expressive communication, this area of development is considered a priority in established educational intervention for them. There is a wealth of literature reporting research into the development of communication in normal and developmentally disabled children, some of which addresses issues related to intentional or lack of intentional communication in the population under investigation in this study. It seems that formal ways of learning might not always be appropriate for the population with developmental disabilities as a group and Rett syndrome population in particular. The fact that there might be important novel forms of learning, that we can not assess at this point, urge us to look for alternative ways of learning for this population (Demeter, 2000). The next section will address this literature in order to provide a reference point from current thinking to relate to in determining communicative potential in girls with Rett syndrome.
1.2 COMMUNICATION

INTRODUCTION

The difficulty in regaining speech in Rett syndrome may be due to dyspraxia (Sigafoos & Woodyatt, 1996). The lack of verbal communication skills have set a handful of scientists on a quest for evaluating and researching communication in this syndrome (Sigafoos et al., 2000; Van Acker, 1996; Woodyatt & Ozanne, 1992, 1994). This chapter will consider current knowledge on communication in the population of developmental disabilities, will set a distinction between the description and definition of non-verbal communication, with a focus on up to date research regarding Rett syndrome.

In the literature, the wide and complex field of communication has been defined in a variety of ways, depending on reference to normal or abnormal development. Some typical examples of definition include:

- "The process by which meanings are exchanged between individuals through a common system of symbols." (Webster, 1976).
• "The process of exchanging information by encoding, transmitting and decoding some type of signal during interpersonal interaction (Wilcox, Hadley, & Ashland in Hanson, 1996 p.367).

• Communication is a social process that involves reciprocal interaction (Siegel-Causey & Bashinski, 1997).

• Wilcox et al. (in Hanson 1996) described that the communication for the non-verbal population is when the nonlinguistic person looks at the partner’s face or orients their body towards a partner. Expressive movements such as kicking (to indicate excitement) and facial expression such as smiles for pleasure and frown for discomfort (to indicate emotional state) are forms of communication.

1.2.1 Communication of the normally developed child

The process by which the development of communication occurs in the first two years of life is conceptualized and understood in different ways, according to various theories:

The behaviorist theory takes the ‘nurture’ position and views language as a behavior controlled by antecedent and consequent stimuli thus learned through early experience with linguistic and nonlinguistic stimuli in the young child's environment (Sameroff, 1975).

The transformational theory takes the "nature" position by Chomsky and talks about the ability to acquire language as an innate humane neurological capability. Language development is a biological, maturational process with little affect by the quality of the child's learning environment (Chomsky in Sameroff, 1975).
The social-interactional theory by Stern sees the social interaction between the infant and their caregiver from the earliest age of infancy as the primary origin of the development of communication (Stern, 1974). A strong base for communication development is the impulses borrowed from other people - intersubjectivity. Intersubjectivity is a theory of how human minds "… recognize one another's impulses, intuitively, even without cognitive or symbolic elaboration" (Trevarthen, 1998, p.17), thus enabling us to accept a primary form of interaction as communication.

There are growing evidence that communication motives are supported by the systems of ‘sympathy neurons’ in the regulatory core of the brain (Trevarthen, 1999; Trevarthen and Malloch, 2000).

Aitken and Trevarthen (1997) believe that the newborn infant seeks “…to enter into regulated engagements with subjective processes in other human beings” (p.654). This process begins early, during prelinguistic experience and proves the social-interactive capabilities that are inherent in the infant. The role of timing in human communication is responsible for exchange of “temporal information segments” when people interact with one another (Trevarthen and Malloch, 2000). Pulse, quality of pitch contour and narrative (phases of expression formed by pulse and pitch) are inherent in the healthy parent/infant interaction and the combination of these elements is called communicative musicality (Malloch, 2000; Trevarthen and Malloch, 2000).

The transactional model by Sameroff (1975) sees the development of language as continual and progressive interplay between the organism and its environment, thus incorporating nature and nurture into a combined continuity.

Stage I (0-3 mo) - Reactive Perlocutionary Communication
The infants' form of communication is considered a reflexive reaction period. These reactions include responses to internal and external stimuli such as crying in response to a hungry state (ex. cry - may be interpreted as a signal of distress). The infant does not intend for their behaviors to send a message, although the caregiver perceives the behavior and attributes a meaning (McLean, 1990; Wilcox et al., in Hanson, 1996).

This stage is used to describe pre-intentional acts that function to have communication effects.
**Stage II (2/3-8/9 mo) - Proactive Perlocutionary Communication** (also defined as pre-intentional communication).

During this period, behaviors produced by the infants are without a purpose, but have a meaning (attribution of meaning) to the person in the surrounding environment.

Communication occurs because interactive partners attribute meaning to the infant's behavior. During the pre-intentional stage, the task of the interactive partner, is to perceive the behavior, attribute a contextually appropriate meaning and then respond (Iacono, Carter, & Hook, 1998; McLean, 1990; McLean & Snyder-McLean, 1987; Wilcox et al., in Hanson, 1996).

The role of the caregiver is of utter importance as it takes a form of regulating the infant's communication (Aitken & Trevarthen, 1997).

The infant's purposeful actions on his environment serve to signal her/his affective states, desires, interests, likes and dislikes to responsive caregiver. The infant's comprehension is in recognizing and responding to ritualized language in familiar routines. His/her production (expression) is by discovering the ability to make things happen through indirect means (McLean, 1990).

The typically developing infant acquires informal communication skills before the emergence of symbolic language. They develop communicative competence in the perlocutionary stage (6 months) when many of the infant's informal and idiosyncratic behaviors (vocalization, facial expressions, body movements, eye gaze and gestures), are interpreted by the caregiver as if the infant is communicating. As a result of the caregiver's contingent responses to the actions, it is believed that these behaviors progress to the illocutionary stage during the second half of the first year.

In this stage, perlocutionary acts have been shaped into intentional forms of communication. (McLean & Snyder-McLean, 1987). Intentional communication consequently emerges.
Stage III (8/9-12/15 mo) - Emerging Illocutionary Communication (also known as the intentional communication stage).

The infant shows deliberate attention to an interactive partner. An infant will actively seek the attention and response of a partner through the use of vocal and or gesture behaviors such as: shouting to seek attention (Iacono et al., 1998; McLean, 1990; McLean & Snyder-McLean, 1987; Wilcox et al. in Hanson, 1996).

Intentional communication act used in several studies looks at it as "an event in which the child directed a motor and/or vocal act toward the adult and awaited a response from the adult" (Wetherby et al., 1988, p. 241). An intentional act is if the person directs the behavior(s) towards an adult and shows evidence of the desire to achieve a goal (for example through persistence of the behavior, ceasing the behavior on obtaining the goal or providing some other form of satisfaction). These can be obtained through modalities such as: vocalization, eye-gaze, gestures, or smiles towards the adult (Iacono et al., 1998).

In the area of comprehension, the infant recognizes and responds to some single words. The infant's production (expression) is in approximates gestures, inflectional pattern and phonological systems of mature speaker to communicate a variety of intents. The child uses objects to have an effect on other people and can use people to have effect on an object (McLean, 1990). It is no wonder that individuals with Rett syndrome are believed to have not reached this stage. Due to functional disabilities such as apraxia and the limitation of hand use, there are limited ways for them to convey intentional communication.

At this stage the child expects response to her/his communicative signals and, with increasing experience, becomes more and more persistent if these signals are not responded to (McLean, 1990). This is exactly where the individual with Rett syndrome fails. She has no way to indicate communicative signals, to be understood, and to repeat experiences.
Around 12 months, the child begins to acquire his first words and enters the fourth stage of language development (the locutionary stage), which is associated with the ability to acquire symbolic communicative acts (e.g. speech and formal gestures) (McLean, 1990).

**Stage IV (12/15-18/24 mo) - Conventional Illocutionary and Emerging Locutionary Communication**

During his second year, the typically developing child will progress from prelinguistic to linguistic communication. They will use conventional and distal gestures paired with appropriate intonated vocalizations, to communicate with a wide variety of intentions. Proto-words and words increase.

The child's comprehension is in recognizing and responding to many single words and logical multi-words combinations. There is a functional importance of early communication acts in the development of language as it promotes language competence (McLean, 1990).

As for the child with normal development, despite small differences between researchers and specialists, and some differences in definitions, there seem to be an agreement on the over all stages of the development of communication. When dealing with children with developmental disabilities there are many differences between populations, pathologies and syndromes that make it unrealistic to search for any commonalties in their development of communication.

**1.2.2 Communication of the developmentally impaired child**

The present section will discuss the following points:
1. What does the term severe communication impairment mean?
2. Is the use of conventional terms appropriate for the population with severe communication impairment?
3. What are the appropriate terms for communication with the population of severe communication impairment?

4. What is the appropriate framework for the population of severe communication impairment?

5. How does Augmentative and Alternative Communication [AAC] enhance communication for the population of severe communication impairment?

1. What does the term severe communication impairment mean?

Over the years there have been repeated reports from professionals concerned with the development of communication in individuals with developmental disabilities, arguing that normal communication development is inadequate for this population (Iacono et al., 1998; McLean & McLean, 1993; Siegel-Causey & Bashinski, 1997).

Extensive publications in the area of communication emerged from the considerable importance of communication in this population, as these individuals do not acquire functional spoken language. They typically develop a range of abnormal, possibly intentional communication and limited comprehension of speech is sometimes present (Iacono et al., 1998; McLean & McLean, 1993; Siegel-Causey & Bashinski, 1997).

They may also exhibit accompanying disabilities, which include, though not limited to: Epilepsy, physical and cognitive impairment or maladaptive behaviors (Sevcik, Romski, & Adamson, 1999). The term severe communication impairment means that the individual's speech is "... permanently inadequate to meet his or her communication needs and whose inability to speak is not due primarily to a hearing impairment" (Asha, 1981, p.286).
2. Is the use of conventional terms appropriate for the population with severe communication impairment?

Traditionally, the primary focus of communication intervention for the developmentally disabled population was to enhance speech as well as to develop prerequisite skills believed necessary for the emergence of verbal language. Throughout the years, many contributors expanded the scope of communication and language intervention from its primary focus on the expressive aspects of communication (learning to talk) to a broader emphasis on the multiple process of communication, including receptive communication (expanding comprehension) (Iacono et al., 1998; Siegel-Causey & Bashinski, 1997; Sigafoos et al., 2000).

Guidelines for identifying pre-intentional communication acts are not evident in the literature, since by their very nature, these are defined according to an adult's subjective inferring of intent to any signal elicited by a pre-linguistic child. Reliability for pre-intentional acts has not been reported (Iacono et al., 1998).

It is important to determine intentional communication skills in a population that may not develop language skills in order to facilitate the implementation of non-verbal communication. In a study by Iacono et al. (1998) some participants with severely developmental disabilities, despite physical limitations, showed some minimal emerging intentional communication in accordance with the published criteria for intentional communication acts. They found that the application of published operational criteria for intentional communicative acts underestimate the ability of the severely impaired population. The important criterion evident in many definitions of coordinated orientation between object and person was missing in most of the communicative acts observed. Yet, it is this behavior that may be the most difficult for this population to demonstrate. Sigafoos et al. (2000) proposes that it is unrealistic to use the terms ‘non-symbolic’, ‘pre-symbolic’ or ‘pre-linguistic communication’ when describing communication skills in the developmentally disabled person. These terms belong to communication skills in
normally developed individuals and cannot be so easily transferred and applied to the population with developmental disabilities.

3. What are the appropriate terms for communication with the population of severe communication impairment?

It is important to find the right definition for levels of communication for the severely disabled individuals so that overestimation or underestimation of his/her communicative level will not occur. Overestimating the child's skills may result in communicating on a level (a symbolic level for example), which may result in failure and the reducing of his/her attempts to communicate. Underestimating the child's skills may result in denying him/her accesses to a symbolic system of communication, thereby causing communication frustration and forcing the child to continue relying on non-symbolic signals (Iacono et al., 1998).

Given the developmental importance of perlocutionary and illocutionary acts, Sigafoos et al. (1999, 2000) urge the investigation of communication potential of children with developmental disabilities. Iacono et al. (1998) found in their investigation that children with intellectual, physical and sensory impairments rarely demonstrated co-ordinate attention, considered the clearest indicator of intentional communication. This may be the result of the children’s lack of clarity in signaling the intention of communicative behaviors and this has serious implications for the assessment and provision of appropriate intervention for individuals with severe and multiple disabilities.

‘Potential Communicative Act’ is a term suggested by Sigafoos et al. (2000). This term acknowledges the possibility that existing informal and idiosyncratic behaviors could become effective forms of communication. It also acknowledges that some behaviors may be symbolic (e.g. manual sign, pointing to pictures on a
communication board). In addition, the use of this term avoids the issue as to whether these actions do in fact represent ‘true’ (intentional) communication.

4. What is the appropriate framework for the population of severe communication impairment?
Very little attention is paid to the partner or directed toward the individual's communication needs in natural contexts (Siegel-Causey & Bashinski, 1997). Interventions that focus strictly on the learner have begun to be replaced by naturalistic interventions and incorporate joint action routines. Siegel-Causey & Bashinski, (1997) suggest a ‘Tri-Focus Framework’ incorporating a partner, learner and environmental context in which the individual's communication interaction takes place. They offer a new way when working on communication with a population with no apparent symbolic communication or individuals with emerging of intentional communication. They look at six primary influences that form the foundation for the ‘Tri-Focus Framework’. These include special education, severe disabilities, augmentative communication, speech-language, psychology, and infant development.

Developmentally disabled individuals share the need for extensive and ongoing support in order to participate in home, school, and community activities. These individuals typically learn slowly and often fail to notice relevant features of what is being taught, do not demonstrate learned skills spontaneously, and do not generalize to new situations (Siegel-Causey & Bashinski, 1997). It is important for the caregiver to detect and recognize when the child is communicating and then to make sense of what the child is trying to convey (Trevarthen & Burford, 1995).

5. How does AAC enhance communication in the population of severe communication impairment?
There are many types of communication modes in the AAC, from manual signs and communication boards to speech-output communication devices (Sevcik, Romski, & Adamson, 1999) and these are valuable for the persons with developmental
disabilities (Calculator, 1999). For the individual with severe communication impairment the use of different types of alternative communication devices is a technical solution, and the importance of communication is much more than just the ability to express one’s thoughts and desires. Communication is more than achieving a set of skills - it is about "taking one's place in the world of humans..." (Trevarthen & Burford, 1995, p. 147). The act of communication starts after a human being is able to define to himself his basic wants and needs. By incorporating AAC into the life of the developmental impaired person, the opportunity to communicate with their surrounding indicating their choices and preferences can be achieved. Therefore the capacity to make and indicate choice through some means of communication is considered a primary and important function for all human beings.

1.2.3 Preference and choice

Preference and choice as a concept and value are embodied in the principal of normalization, empowerment, quality of life and self-determination (Hughes, Pitkin, & Lorden, 1998). Choice making is a right, and for most people, a cherished component of life (Bambara, Koger, Katzer, & Davenport, 1995). The freedom to choose should not be approached as a programmatic end in itself, the opportunity to give preference and choice typically are viewed as critical to the process of one's personal growth and fulfillment (Hughes et al., 1998).

Despite philosophical and empirical support for the critical role of preference and choice in improving one's performance, life style, and day-to-day experiences, numerous studies indicate that individuals with severe disabilities have limited opportunities to express preferences or exercise choice in their daily lives as compared to the persons without disabilities (Hughes et al., 1998; Sigafoos, Laurie & Pennell, 1995, 1996). In recent years, the value of choice making for persons
with severe disabilities has been discussed repeatedly in terms of personal autonomy and dignity, which are essential to one's quality of life (Hughes et al., 1998; Nozaki & Mochizuki, 1995; Sigafoos et al., 1995, 1996). Choice making is a skill that can provide motivation for achieving a functional level of communication, and create and enhance meaningful experiences for children with the most severe disability (Sigafoos et al., 1995).

Unfortunately, in the lives of people with severe disabilities, choice-making opportunities have been noticeably absent (Bambara et al., 1995). The absence of choice making can have a devastating effect on individual’s quality of life and emotional development. Limiting the opportunity to control the environment can place an individual at risk for developing learnt helplessness and as result develop behavioral excesses (Bambara et al., 1995; Guess, Benson, & Siegel-Causey, 1985).

Several explanations exist for the lack of choice making among individuals with severe disabilities.

1. The individual lacks the skills needed to communicate their preference clearly.
2. Caregivers may not consider that the individual is capable of intentional choice making, and appropriate assessment to determine potential has not been undertaken.
3. Caregivers may fear that the individual may make poor or indiscriminate choices.
4. The choices may interfere with the caregivers' instruction goals. (Bambara et al., 1995; Guess et al., 1985).

A number of studies have shown that when individuals with severe disabilities are provided with a choice between tasks, participation increases in vocational (Bambara, Ager, & Koger, 1994), leisure (Dattilo & Rush, 1985), and self-care (Rice & Nelson, 1988) activities. A link has also been established between choice and the prevention of behavioral problems (Dyer, Dunlap, & Winterling, 1990; Bambara et al., 1995).
Choice making was proven to increase motivation, task initiation and eliminate aggression in a case study performed with a person with severe disability (Bambara et al., 1995). In the same study it was noted that positive outcome such as smiles, happiness, jokes with others in the surrounding appeared as a result of choice making. The individual initiated more tasks than expected (Bambara et al., 1995). The ability to request enables the individual to express wants and needs and to gain control over interactions with others (Sigafoos et al., 1996). Structured choice making opportunities might be used to assess preferences and to identify reinforcers (Sigafoos et al., 1995; Bambara et al., 1995). Choice making therefore relies on the development of communication to the degree that intention and need can be recognized and understood.

"Though techniques can be employed to improve the lines of communication, the power of the relationship itself should not be forgotten. Communication is more than achieving a set of skills - it is ... a shared emotional understanding between individuals” (Trevarthen & Burford, 1995, p. 147).

From reviewing current literature, it is quite evident that on top of lacking basic communication abilities due to their primary limitation, individuals with severe communication impairment are handicapped by the lack of sufficient opportunities for preferences and choice making. These opportunities have been noticeably absent. Individuals with Rett syndrome are presented with impaired communication. What might be expected from them in terms of communication abilities? How well motivated are they to acquire sufficient communication strategies? Will they reliably exercise them when presented with opportunities to communicate?
1.2.4 Communication in individuals with Rett syndrome

A thorough review of the literature to explore communication in individuals with Rett syndrome and the structure has been adopted to follow a logical process of investigation:

- Cognitive and communicative abilities in individuals with Rett syndrome.
- The basic fault causing communication loss in individuals with Rett syndrome.
- The inability to regain lost communication in individuals with Rett syndrome.
- Reports describing communication in individuals with Rett syndrome as pre-intentional.
- Reports describing communication in individuals with Rett syndrome as intentional.
- Indirect ways to reveal communication skills in individuals with Rett syndrome.
- Clinical suggestions to enhance communication skills in individuals with Rett syndrome.
- A look into the future.

1.2.4.1 Cognitive and communicative abilities in individuals with Rett syndrome.

Individuals with Rett syndrome have been characterized as having severe communication impairment. Although they are diagnosed as severely developmentally disabled, it is still difficult to assess their intellectual potentials, and more so, their communicative abilities through conventional tests of cognitive
and language ability. In order to assess cognitive ability through standardized tests, one is required to have either expressive language or the ability to use hands functionally. Individuals with Rett syndrome have severe impairments in both areas of function, and therefore no tests thus far have been adapted to their needs (Van Acker, 1996; Weiss, 1996). Demeter (2000) suggests that in order to get a more complete image of their information processing capacity, one should look into the girls’ interests. Nevertheless, it has been suggested by some that many girls have a high level of understanding and respond appropriately to different situations and events (Budden, Demeter, 2000; Meek, & Henigham, 1990; Hunter, 1999; Lindberg, 1991; Lewis & Wilson, 1996). While Woodyatt and Ozanne (1994) mention fluctuating attention and lack of motivation, Watson, Umansky, Marcy, & Repacholi, (in Demeter, 2000) demonstrated that girls with Rett syndrome have the capacity for showing intention and preference concerning interesting stimuli.

The same dichotomy seen in regard to cognitive abilities is found when trying to locate the Rett syndrome population’s communicative potential. In some studies, parents report normal pre-linguistic behavior in their daughters until the onset of regression (Budden et al., 1990), while other studies argue that early development of communication is already impaired before the girls enter first or second stage (Kerr, 1992). The girls lose significant communication skills, more in verbal expression than in language comprehension (Budden et al., 1990). These findings are supported by a survey undertaken on girls with Rett syndrome measuring communication skills at a Research Institute in the USA. The girls' expressive language was found to be less developed than their receptive capabilities (Lewis &Wilson, 1996).
1.2.4.2 The basic fault causing communication loss in individuals with Rett syndrome.

Reasons for loss of receptive and expressive language resulting in severe communicative impairment is unclear, although some studies have suggested various explanations:

1) Girls with Rett syndrome lose expressive and receptive language because of impairment in motor control for the development of speech (Fontanesi & Haas, 1988; Lindberg, 1991; Witt Engerstrom, 1990). This is consistent with motor control problems in this population, such as loss of functional hand use and loss of mobility, and the presence of involuntary lip and tongue movements (Sigafoos et al., 1996).

2) Due to severe and profound cognitive impairment associated with Rett syndrome, the loss of language could be as a result of stagnation at the pre-intentional stage of language development (Snyder-McLean et al., 1988).

3) Lack of motivation to interact and poor mobility may be more likely why the girls score very low when it comes to communicating (Woodyatt & Ozanne, 1992, 1994).

1.2.4.3 The inability to regain lost communication in individuals with Rett syndrome.

There have been two explanations for the failure to regain speech after the regression in individuals with Rett syndrome. Sigafoos et al. (1991) relates to the physical difficulties suffered by girls with Rett syndrome, which include dyspraxia,
a deficit of motor planning and motor learning which results in difficulty of acting in and on the environment and is often viewed as an impairment of the intentional motor act. This is evident mainly in the lack of purposeful hand movement of Rett syndrome (Witt-Engerstrom, 1990; Lindberg, 1991).

The second explanation reported by Woodyatt & Ozanne (1992, 1993, 1994) is their severe cognitive impairment, which may result in their lack of intentionality (motivation) in communication. This lack of intentional communication and the stagnation during the pre-intentional state has been explained as the lack of verbal skills in this population (Snyder-McLean et al., 1988). Severe dyspraxia and severe cognitive disability are now the accepted explanations for the failure of individuals with Rett syndrome to regain speech (Woodyatt & Ozanne, 1992, 1994). If loss of speech stemmed from dyspraxia alone, then one would expect that individuals with Rett syndrome may retain communicative intent (Sigafoos et al., 1996).

1.2.4.4 Reports describing communication in individuals with Rett syndrome as pre-intentional

Woodyatt & Ozanne (1994) studied communication behaviors in girls with Rett syndrome and found that their lack of skills is consistent with the girls' profound disabilities. The girls function at the pre-intentional level of communication. It is suggested that both physical and cognitive factors effect the communication level (Woodyatt & Ozanne, 1992, 1993, 1994). Other findings of a similar nature can be found in von Tetzchner’s study (1997), that found little evidence of intentional communicative behaviors among a sample of 42 Norwegian girls and women with Rett syndrome. However, von Tetzchner’s findings were based on interviews with parents.
Woodyatt & Ozanne (1994) observed four girls with Rett syndrome during attempts at intentional communication using words that seemed to have been retained prior to their developmental regression. Only one girl out of the four indicated some intentional communication. If language development regresses to a pre-intentional stage, then the non-symbolic actions may not represent intentional communication response (Budden, 1990; Woodyatt & Ozanne, 1994). The studies talk about the girls' total lack of imitation behaviors, which contraindicate communication development (Woodyatt, & Ozanne, 1992). Trevarthen & Burford (2001) believe that the girls appear to retain an accessible capacity for an infancy level of communication. Although non-symbolic acts such as eye contact, facial expression, body gestures and vocalization are sometimes used as communication acts, it is unclear if these actions represent intentional communication in individuals with Rett syndrome (Sigafoos & Woodyatt, 1996). Although caregivers interpret and attach communicative meaning to the girls' vocal and non-verbal behaviors, it had been found to be at the pre-intentional level (Woodyatt & Ozanne, 1992, 1993, 1994). Demeter (2000) summarizes this topic by writing that "... most authors judge the abilities of the RS girls to be equal to those of infants" (p. 227).

More comprehensive assessment has been recommended to determine functional and intentional levels of communication in individual with Rett syndrome. A rather pessimistic description of communication potentials in the Rett syndrome population is summed up by Woodyatt & Ozanne (1992): "Unless intentionality is present, any intervention aimed at increasing output (such as learning through a symbol system)... will not be used as a functional communication system but rather will be inconsistently produced" (p. 171).

‘Choice making’ was examined in seven girls with Rett syndrome for the purpose of identifying intentional communication (Sigafoos, Laurie, & Pennell, 1995). The girls were given opportunities to choose between two items (such as: food, drink, and play activities) during structured choice making activities. Evidence showed that none of the girls ever refused an item given to them even after they had not
In addition, when deeper analysis was made, it appeared that the girls in their study had very few definite preferences for the two offered items (Sigafoos et al., 1995). In contradiction to these findings, a music therapy survey in girls with Rett syndrome found that passive acceptance of all types of music did not exist. This survey found that the girls had definite musical preferences (Holdsworth, 1999). Such differences in findings are probably due to the child's difficulties in establishing interest in certain objects and in contrast the strength they show in focusing on auditory information (Demeter, 2000).

In a recent study on storybook interaction, it was noted that girls with Rett syndrome were vocalizing and gesturing during the storybook interaction, but their meaning was unclear to the researcher who suggested that the parents give meaning to their child’s behaviors (Koppenhaver et al., 2001). "This retained capacity for interaction, the girls' greatest strength, should underpin educational and therapeutic strategies to achieve maximum effect” (Trevarthen & Burford, 2001, p. 321).

If these types of actions do not represent intentional communication, then the actions may be motor movement that may or may not be voluntary (Sigafoos & Woodyatt, 1996).

The lack of success in choice making is explained by the following reasons:

- **Not enough** opportunities to establish preference.
- Inability to **discriminate** between two items.
- Neither of the items offered is sufficiently **motivating** to evoke selection or preferences.
1.2.4.5 Reports describing communication in individuals with Rett syndrome as intentional

In contrast to the studies reported above concerning low levels of communication ability in girls with Rett syndrome (Budden, 1990; Woodyatt & Ozanne, 1992, 1993, 1994) there are others (Lindberg, 1991; Sigafoos & Woodyatt, 1996) that report individuals with Rett syndrome as having some intentional communication. Lindberg (1991) has not undertaken experimental studies, however she anecdotally reports some examples from her vast experience in this population. She proposes that “all of the girls understand the meaning of some learned words and associate them with specific objects and situations” (p.54) and believes the girls clearly show some abilities in comprehension.

Sigafoos & Woodyatt (1996) suggest that: “One might expect children with Rett syndrome to retain communicative intent. This intent could be manifested in non-symbolic act, such as eye contact, facial expression” (p. 23). Watson et al. (1996) in regards to social stimulus demonstrated intent and preference in the case of one girl with Rett syndrome.

Can the lack of engagement in experimental activities by children with physical and cognitive impairment be linked to lack of motivation and inattention to stimuli? Is it possible that experimental situations did not present the children with expressive communication that was functional, age appropriate and motivating, causing the girls to refuse in taking part in communicating? Dr. Kerr (1992) suggests the possibility that individuals with Rett syndrome produce their best results when no demands are made (which is not the case in research settings), but when an appropriate opportunity is available. Such a remark might explain the differences between clinicians’ experiences of the potential of females with Rett syndrome and researchers’ findings, as concerns the communication abilities in Rett syndrome.
1.2.4.6 Indirect ways to reveal communication skills in individuals with Rett syndrome

Lewis & Wilson (1996) describe the individual with Rett syndrome as wanting to express their feelings, but have difficulties in communicating these feelings and also in demonstrating their intellectual capabilities. Is it possible that previous studies were unable to overcome such difficulties or find participants that “fit into their frame of reference and their system of values” (Lindberg, 1991. p.54), and subsequently obtain results that showed the individuals lack of interest as reflecting her lack of intellectual or communicative abilities? Or maybe their communication through idiosyncratic movements seem meaningless to the untrained eye (Trevarthen & Burford, 2001)?

The girls are able to initiate some form of communication in different settings (Budden et al., 1990). The period after regression is a time when communication can improve and the girls are seen making effort in communicating through their eye contact (Witt Engerstrom, 1990; Leonard et al., 2001). More typically, they communicate by using non-symbolic means such as gestures, vocalizations and body positioning (Coleman et.al., 1988), and by retaining positive contacts to people's faces/eyes reacting with smiles (Burford & Trevarthen, 1997; Fontenasi & Haas, 1988).

The girls may respond by accurately looking at an object or walking and standing in close proximity to the object. For example, if the girl wants to go out of the classroom, she may approach the door, stand in front of it, either rock, stare or knock with her hand to indicate her wishes. If she is disinterested or bored, she may close her eyes or move away. Some parents use this level of communication to achieve a more structured communicative ambience.

In a case study by Sullivan, Laverick, & Lewis (1995) one girl gained control when using switch-activated toys. Initially she was taught to use these and as she gained
experience she developed independence and initiation in activating the switches in
the classroom. Sigafos et al. (1999) found in their study for assessing behaviors in
three girls with Rett syndrome, that some of the girls’ motor movements and other
idiosyncratic behaviors might have been conditioned as unconventional form of
communication. They called these behaviors ‘potential communicative acts’. These
identified behaviors became evident as a result of three factors: (1) The researchers
interviewed the girls’ staff who reported these behaviors, (2) they observed these
behaviors under conditions of high and low social interaction, and (3) they
observed these behaviors during structured communication probes. The main
findings of the study demonstrated that although girls with Rett syndrome have
limited behavioral repertoires, all had some types of behaviors that were interpreted
by the staff as a form of communication.

The following behaviors were observed in their study and viewed as ‘potential
communicative acts’ (Sigafos et al., 1999):

- Stereotyped hand movements (wringing, rubbing, or clasping of
  hands)
- Eye gaze (looking at staff person for at least 3 sec.)
- Hyperventilation (rapid audible breathing)
- Vocalization (any vocalization other than breathing).
- Facial expression (smiling or frowning)
- Body movement (wiggle, kick, moving head or torso towards or
  away).

They went on and developed an inventory based on the ‘potential communicative
acts’ of children with developmental disabilities and severe communication
impairment. This inventory included eight girls with Rett syndrome and was called
‘Identifying Potential Communicative Acts (IPCA) in Children with
Developmental and Physical Disabilities’ (Sigafos et al. 2000). In the IPCA, they
explained in more detailed communicative behaviors derived from the children
with developmental disabilities (including girls with Rett syndrome), but
From anecdotal evidence, it appears that some individuals with Rett syndrome retain the ability to communicate via non-symbolic behaviors (Demeter, 2000). It would be essential to strengthen and maintain those existing skills and to utilize them within the context of functional communication interaction. Sigafoos et al. (1996) suggest to assess the non-symbolic communication behaviors and to strengthen them by responding to the child's initiations. In a later study, Sigafoos et al. (1999) assessed five communication functions during structured communication probes. These included: Greeting, conversation, requesting object, protesting and requesting more. Stereotyped hand movements and eye gaze were relatively high when the girls were engaging in the structured probe activities. One girl’s hyperventilation was high and the two others were low. Vocalization was quite low in two of the girls and one girl vocalized more during ’more’ activities. All exhibited high levels of body movement during structured activities of ’request’, ‘protest’ and ‘more’.

Facial expression was quite low in all girls except in one girl following ‘request’ and ’more’ probes. The type of facial expression was undefined. Woodyatt and Ozanne (1994) reported one girl in their study to smile and to express an increase level of activity when asked whether she was interested in hearing a tape with music or stories, then she walked towards the source of the sound. This was a clear indication of communicative response to a question and the acting upon it. In their study, Sigafoos et al. (1999) reported of one girl who moved forwards, towards a desired item during the ‘more’ trials. They saw this trend of body movement forward as an unconventional form of requesting. Some girls communicate by approaching and touching another person to gain attention, or push away a person when they do not wish to be engaged (Lindberg, 1991; Lewis & Wilson, 1996).
The girls seem to indicate likes and dislikes and feelings such as happiness and sadness. Girls with Rett syndrome often laugh, make loud sounds that seem to express happiness, or hug and kiss a familiar person to express affection. Humorous interaction may be perceived as a form of communication. Wigram is described in Burford and Trevarthen (1997) as interacting with a girl with Rett syndrome in a humorous and playful manner. All of these behaviors indicate the need to promote their communication, to provide the opportunity to express their strong desires and emotions and to help them take their "place in the world of humans" (Trevarthen & Burford, 1995, p. 147).

Because many of the individuals with Rett syndrome appear to use behaviors such as eye contact as mean of functional communication, it should be incorporated into a formal system of communication such as the usage of graphic system in Augmentative and Alternative Communication. In Lindberg's study (1991) she reported that females with Rett syndrome show great interest in pictures. Many of the girls in her study associated a picture with the real object and many interpreted new and unfamiliar pictures. Besides the act of recognizing the pictures with objects, it is reported that some girls are able to make choices (Sigafoos et al., 1995, 1996). This ability to make choices and to request enable the child to express wants and needs, and as a result to gain control over the interaction with others.

As Rett syndrome is considered a neuro-developmental disorder (rather than neuro-degenerative disease as it was once thought), it is believed that this population may have the capacity to learn new skills (Demeter, 2000; Jacobsen et al., 2001; Leonard et al., 2001) if given the opportunity, even after reaching adult life (Demeter, 2000; Kerr, 1992). Individuals with Rett syndrome can identify symbols when taught in a motivating form. This was demonstrated by a study that examined
the effectiveness of computer-based interactive language development system (Hetzroni & Rubin, 1998).

1.2.4.7 Clinical suggestions to enhance communication in individuals with Rett syndrome

Although the individual with Rett syndrome is extremely limited in expressing herself verbally, she has been found in the past (although not consistently), to be able to recognize and respond to single words and multi-word combination (Lindberg, 1991). If we consider suggestions and insights made by researchers in communication for Rett syndrome we find the following:

1. Initially, communication should be on a form of non-linguistic interactive level (Burford & Trevarthen, 1997). People with Rett syndrome should be given opportunities to learn and communicate so that they could achieve greater influence over their immediate environment (Demeter, 2000).

2. The intervention should initially be undertaken through highly motivating activities (Burford & Trevarthen, 1997; Elefant & Lotan, 1998).

3. Communication experiences should "look to the interests of these girls and use them" in order to get intention and preference (Demeter, 2000, p. 230).

4. It is important to provide opportunities in making their own choice in order to adjust their environment according to their own needs (Kerr, 1992).
5. They can participate in and benefit from **structured demand-based interactions using aided communication** (Sigafoos et al., 1996).

6. They have **a need for Augmentative and Alternative Communication** due to the loss of their speech (Sigafoos et al., 1996).

7. They can benefit from **learning symbolic requests** (Sigafoos et al., 1996; Sigafoos & Woodyatt, 1996).

8. They can benefit from **structured communication opportunities** (Sigafoos et al., 1995).

### 1.2.4.8 A look into the future

Further research is needed to develop more effective interventions for individuals with Rett syndrome (Sigafoos et al., 1996). Burford and Trevarthen (1997) imply that a goal-directed program may not work for the girls as a more intensive interaction teaching based on early mother-infant interaction. "This approach reveals the value of the communicative relationship in developing the child's abilities to initiate, take turn and understand cause and effect" (p.4). Demeter (2000) proposes learning through basic form of learning such as conditioning and believes that the child will learn more through associative learning.

While the focus on teaching communication skills is important, it is equally important to identify variables that may improve the general communicative responsiveness in children with developmental disabilities. Identifying interventions can create more effective opportunities to communicate and strengthen the existing communication skills in individuals with Rett syndrome (Braithwaite & Sigafoos, 1998), thus elevating their quality of life.
Individuals with Rett syndrome show a strong desire to communicate. This is evident by their eye gaze, facial expression and body gestures. Due to their interest to communicate with others, this desire should be pursued. Educational and therapeutic needs for individuals with Rett syndrome should not be any different than those for other children with multiple disabilities who have needs in basic skill areas such as: communication, social skills, daily living care and gross and fine motor skills (Sigafoos et al., 1996).

During my experiences when working with girls with Rett syndrome, I have seen their high need for motivating activities. They need objects and materials they like and prefer. Once they find an interesting and motivating environment, they are likely to become fully involved, attuned and able to learn. Music is reported to be very motivating as a stimulus, and it is therefore natural it should be utilized as a medium for promoting communication for individuals with Rett syndrome. When using music to promote communication, we could be able to observe communicative behaviors that will allow access to preferred objects and activities, and enhance control over the child’s environment.
1.3 MUSIC THERAPY

1.3.1 Music therapy with the developmental disabled children

Children with developmental disabilities are the most frequently treated population in music therapy (Hanser, 1999). The literature is abundant with different types of interventions in music therapy for the developmental disabled. It looks at individual or group music therapy and structured vs. open improvised music therapy (Aigen, 1995; Alvin, 1976; Boxill, 1985; Bruscia, 1987; Grant, 1989; Nordoff & Robbins, 1971, 1977; Oldfield, 1995; Robbins & Robbins, 1996; Voigt, 1999; Wigram, 1995).

1.3.1.1 The power of music therapy with developmental disabilities

Music therapy may increase general levels of responsiveness and engagement in children with disabilities (Toolan, & Coleman, 1994). It aims at bypassing the areas of pathologies and accesses the healthy ones. Practitioners claim that cognitive and affective processes not otherwise detected are activated through music, open new channels and promote personal development (Aigen, 1995; Boxill, 1985; Hill, 1997; Nordoff & Robbins, 1971, 1977).

Nordoff and Robbins worked with developmentally disabled children through an improvisational music therapy approach called “Creative Music Therapy” and looked at musical sensitivities inherent in this population (Nordoff & Robbins, 1977). The disabled child is called “condition child” as he has come to be with his
neurological and physical deficits. His potential for development has not yet been released (Robbins & Robbins, 1996).

Nordoff & Robbins (1977) believe in the intelligence these children reveal during musical interactions. Each child, no matter how impaired, is a "music child" and is believed to possess innate musicality. The severely impaired child may initially respond in a reflexive form and only after some direction and guidance of the therapist who stimulates the child (rather than observes and follows him), the child may begin to free and open his "music child" (Oldfield, 1995).

Other approaches view the developmentally disabled child from a holistic perspective and therefore, enhancing motor, communicative, cognitive, affective and social skills through music therapy should not be considered in isolation during the therapeutic process, but rather taken into consideration at the same time (Alvin, 1976; Boxill, 1985; Bruscia, 1987; Grant, 1989). The Orff music therapy suggests that individuals with developmental disabilities need to be given opportunities to interact according to their individual abilities, and at the same time given the support in developing these further (Voigt, 1999). Alvin (1976) discusses the interaction of emotional, social, motor and cognitive domains of disabled individuals and how these are activated by music therapy. Musical experiences affect these different areas, and the important goals when working with the developmental disabled are in developing cognitive skills (Alvin, 1976; Bruscia, 1987).

Music is a powerful motivator. Even the most profoundly disabled child responds and cooperates during musical dialogues (Trevarthen, & Burford, 1997). The child becomes not only emotionally involved in the music, but also in his own self-realization (Nordoff, & Robbins, 1977). The feeling of success is a motivating force for learning and may bring to a change and to ground work for intrinsic learning (Boxill, 1985).
1.3.1.2 Music therapy as a promoter of communication

The structural form of music provides security, predictability, organization, encourages spontaneous participation in vocal, movement and participation (Alvin, 1976; Wigram, & Cass, 1996). All of this, paired with a supportive environment may enable the child to enhance learning and to facilitate communication. Individuals with communication disorders indicate their enjoyment to music and sounds and may appear more alert and less isolated when involved in this type of communicative process. Music seems to motivate and interest this population (Oldfield, 1995). The children can have the opportunity to express themselves during music therapy and as a result this could generate feelings of success. The child can gain positive development and self-actualization within the frame of their disabilities (Boxill, 1985; Nordoff & Robbins, 1977; Voigt, 1999).

Braithwaite and Sigafoos (1998) found in their study, that when including communication opportunities within a musical activity it could lead to increased appropriate communication responses for some children with developmental disabilities. They suggest that these children produce higher levels of spontaneous speech during music therapy and it may help stimulate communication more than during structured language activity. The results of their study show that musical antecedents facilitate communication responsiveness in some children with developmental disabilities. It is believed that these motivational factors are functional in generating greater communication responsiveness. The musical antecedent condition enhanced attention and reduces stereotyped movements.

Music therapy can provide opportunities in making choices between different activities during the sessions. Some studies have found that through some type of communication system the individual with developmentally disabilities can expand his/her opportunities in choosing. Results of a study by Nozaki & Mochizuki (1995) found that individuals with profound disability could express distinct musical preferences when given choice-making opportunities.
Using symbols to songs during music therapy with cerebral palsy children was reported to have been successful (Brodsky, 1984; Herman, 1985). These children had learnt and could demonstrate knowledge of song lyrics by using ‘Blyss’ symbols. Self-confidence, requests for specific songs and independence were viewed as these children began to control their environment through the symbols (Brodsky, 1984). Through the symbols the child can enhance communication and interact with his surroundings. Music can set a climate that fosters communication through symbols, helping the non-speaking child to express himself, his ideas, feelings and attitudes (Elefant & Lotan, 1998; Herman, 1985).

Music therapy has varied models, philosophies, approaches and techniques relating to the type or style of music utilized during therapy. There are therapists who predominately use improvised music, while others use pre-composed music. The same therapist may apply both kinds depending on the clients' needs at the time of therapy. For this study pre-composed children's songs were administered.

**1.3.2 Pre-composed children’s songs as part of the treatment of children with developmental disabilities**

The value of pre-composed songs for developmentally disabled children is evident by its wide use in music therapy (Boxill, 1985; Brodsky, 1984; Herman, 1985; Hibben, 1992; Elefant & Lotan, 1998; Levin & Levin, 1975; McLean, 1990; Nordoff & Robbins, 1971; Oldfield, 1995). Using songs (improvisational and pre-composed) in therapy promotes important therapeutic goals such as positive experience, integrating emotional, physiological and interpersonal aspects of the self (Boxill, 1985) as well as communicational and academic goals (Levin & Levin, 1975; Nordoff & Robbins, 1971; Wood, 1975). "Singing can be an experience of arousal for the handicapped child, of freedom from many of the confusions and restrictions of pathology" (Nordoff & Robbins, 1971, p.22).
1.3.2.1 Songs as a natural element in music therapy

Using songs for children with developmental disabilities is as natural and appropriate as a mother singing to her infant. It is linguistically simple and repetitive just as ‘motherese’, a form of communication, which does not emphasis on its vocabulary or grammar, but on its intonation or prosody. ‘Motherese’ takes the form of exaggerated intonation and stress patterns in the vocalizations. The melody is the message (Fernald, 1989). Developmentally disabled children do not necessarily respond differentially to the text or grammatical structures of the song's messages, however, they are sensitive to changes in auditory stimuli physiologically and on emotional levels (Boxill, 1985; McLean, 1990).

The use of songs in music therapy for developmental disabled children can take many shapes and forms. Songs can be familiar and unfamiliar, simple or complex, with or without repetition, verbal or non-verbal (Bruscia, 1987). Songs can be used as a tool in individual therapy, or in-group treatment (Wood, 1975).

1.3.2.2 Effects of songs in music therapy

Songs about activities or events that children may be familiar with, also believed to be most effective for developmentally disabled children. These songs can be used to teach different functional skills and concepts (Boxill, 1985; Levin & Levin, 1975; Nordoff & Robbins 1971). Nordoff & Robbins (1968) wrote children’s play songs for group music therapy for developmentally disabled children. Their idea behind the songs was that the children would preserve the element of play or game despite their disabilities. Through the songs the children could relate to one another as well as purely have fun and enjoyment. The content of these songs refers to issues children could relate to and as a result they would enjoy singing and listening to them.
Children's songs in many countries are characterized by simple harmonies, melodies and are easy to remember. Most children's songs have a clear beginning and an ending that is also a source of comfort and satisfaction. They can therefore feel an achievement of a finished product (Oldfield, 1995).

There are elements in a song that establishes the sense of security, reliability, containment and trust. Children can gain a sense of security and safety from the sound and the rhythm associations with familiar songs. The songs are simple, predictable in their rhythmic pattern and have plenty of repetition that is more accessible to children. According to Grinnell (in Bruscia, 1985), the influence of songs in music therapy is not constant. It is described as a developmental therapeutic process that takes place between the child (the client) and the music therapist through the use of songs. The songs are used to facilitate communication and to prepare the child for a personal interaction with the music therapist. Ruud (1998) concludes that: “Musical interaction through songs helps to establish a basic sense of inter-subjectivity through which a child can, from early on, make an impact on another” (p. 60). As the relationship between therapist and client develop, the songs reflect the child’s mood and feeling and promote self-awareness. The therapist uses the same music from one session to the other as it serves as an organizer and gives the session its form (Grinnell in Bruscia, 1985).

In my personal experience during group music therapy for developmentally disabled children I have used pre-composed children’s songs, which have led to emotional and communicative expressions by the children. The children’s responses to the songs were very positive and they requested to hear the songs repeatedly by different communicative forms. Once the children had familiarized themselves with different songs, they were given opportunities to choose between a large song repertoire. In this manner, each child was able to give expression to his personal desires. As a result the child gained control over his environment and events that took place during music therapy sessions. This is a primary experience
that to my understanding uncovers hidden skills and reveals a pathway to expanded intervention of similar nature in other situations.

1.3.2.3 Effects of musical elements in the song

The song and its components (text, form, sound contents, how it is selected and performed) have many therapeutic advantages. The melody (musical sound) is not the only determinant therapeutically (Boxill, 1985; Frank-Schwebel, 1995; Hibben, 1992; Oldfield, 1995). A strong rhythmic pulsation that is repeated over and over in the same tempo and on the same dynamic level can have an effect, which can stabilize both physically and emotionally. Chanting and singing in rhythmic patterns of words can stimulate verbalization and aid in cognitive development (Boxill, 1985).

In the song, there is the principle between the structure and organization and the principle of variability in expressiveness. Structured elements are characterized by the repetition and the ability of the participants to predict what is going to happen in the song. The steady elements are connected to time (tempo, meter, rhythm) as well as tonality, harmony and form. The expressive content is carried by the melody (Aldridge, 1999), range, quality, dynamics and performance (Frank-Schwebel, 1995; Sekeles, 1996). The rise and fall of tones reflect the quality of the text, or the text may reflect the quality of the melodic line (Boxill, 1985).

Rehearsing songs seldom involves exact repetition. There are musical and verbal variations and changes of nuance every time the song is sang (Sekeles, 1996). These small nuances construct an on going discussion between the therapist and the child through songs. Bruscia (1985) speaks of influential musical components such as familiar materials that create comfort and security, whereas unfamiliar songs
stimulate interest. Repetitions provide opportunities to stabilize, where as close ended songs provide structure.

In a study on communication with developmental disabled children, by Braithwaite and Sigafoos (1998), it was reported that improvement in responsiveness and functioning would increase and improve during song antecedent condition and was facilitated by the structure, phrasing, rhythm and lyrics of the songs.

The arguments presented by researchers about the value of songs as a tool in the hands of the music therapist are persuasive. Songs in the treatment of developmentally disabled individuals are natural when incorporated into music therapy procedures. They seem to establish trust, to build structure, to elevate reliability and enhance a sense of security. Different components in the songs may cause specific reaction and changes in the child exposed to them. Such influence will be at the disposal of the researcher as she uses the tool of songs with children with Rett syndrome, for whom music therapy is recommended, even indicated. The literature supporting the effectiveness of music therapy interventions includes various approaches and has proposed the efficacy of improvised music making and song-singing.

1.3.3 Music therapy for individuals with Rett syndrome

Music therapy was recommended by Dr. Andreas Rett as early as 1982 as a mean of treating individuals with Rett syndrome (Rett, 1982). Music therapy evokes positive response in girls and adults with Rett syndrome. Reports have shown how music making promotes and motivates their desire to interact and communicate with their surroundings as well as develops their cognitive, affective, sensori-motor and physical skills (Allan, 1991; Coleman, 1987; Elefant & Lotan, 1998; Hadsell & Coleman, 1988; Hill, 1997; Kerr, 1992; Lindberg, 1991; Merker, Bergstrom-

Over the years individuals with Rett syndrome have been described by the medical world as having severe mental retardation as well as being non-educable (Smith, 1988). Although promoting the use of music therapy for this population, Dr. Rett described them as "practically unable to learn" (Takehisa & Takehisa-Silvestri, n.d.). Other professionals have echoed this view, a belief that to date prevails in some areas around the world (Takehisa & Takehisa - Silvestri, n.d.). Despite these voices, researchers (in different areas of interest) have been applying music (e.g. taped songs and musical toys) when investigating this population (Sigafoos et al., 1996; Sullivan et al., 1995; Woodyatt & Murdoch, 1996; Woodyatt & Ozanne, 1992). In the past few years, reports from different parts of the world have turned the concept that females with Rett syndrome cannot learn (Rettnet, 1998). Some arguments have been presented that music is valuable not only in maintaining the girl's skills but also in stimulating cognitive development and new skills through "channels not yet discovered" (Hill, 1997).

Girls with Rett syndrome’s ability to learn has been stimulated by a number of approaches, i.e. augmentative (non-speech) communication (Elefant & Lotan, 1998), literacy (Koppenhaver et al., 2001), and computer activation (Hetzroni & Rubin, 1998). It is also now evident that they retain words that were used prior to the regression and the question has become how to retrieve that skill (Woodyatt & Ozanne 1994; Zappella, 1992). To date, no reliable means of assessing this population's cognitive abilities has been developed, but attempts are being made to adapt standardized tests for their needs (Demeter, 2000; Van Acker, 1996). However, there is evidence that through music therapy assessment one can obtain relevant information regarding their ability, and their motivation, and such assessments have proven to be successful (Wigram, 1995). "Experienced researchers pleaded for increased research activity on what RS girls are able to
manage, instead of concentrating on their disturbing deficiencies” (Demeter, 2000, p.227). Might music therapy provide such a tool that will be able to adapt and prevail the complex Rett syndrome situation?

Rett syndrome became familiar to the music therapy community only during the last 15 years. Not much was known about their emotional, cognitive and communicative abilities. Lack of knowledge gave way to individual intuitive interventions in music therapy, which have taken different approaches and views (Coleman, 1987; Montague, 1986; Wesecky, 1986). These have changed and expanded as the puzzle of Rett syndrome has unfolded. No matter what approach was taken, there was never any doubt that music therapy promotes a very strong motivational force within individuals with Rett syndrome (Allan, 1991; Coleman, 1987; Elefant & Lotan, 1998; Hadsell & Coleman, 1988; Hill, 1997; Kerr, 1992; Lindberg, 1991; Merker, Bergstrom-Isacsson & Witt Engerstrom, 2001; Montague, 1988; Takehisa & Takehisa - Silvestri, n.d.; Wesecky, 1986; Wigram, 1991, 1995, 1997; Wigram & Cass, 1996).

Some therapists suggested that it was difficult if not impossible for the Rett syndrome population to learn new skills or to re-learn acquired skills. Stereotypic hand movements interfered with any intervention and the purpose of music therapy was to stimulate through the auditory and optical modes (Wesecky, 1986). Ten years later others propose that the Rett syndrome population cannot only re-learn forgotten skills, but are able to learn new once (Takehisa & Takehisa - Silvestri, n.d.).

Some music therapists view music therapy intervention with individuals with Rett syndrome as a way to promote and improve functional skills such as hand use, eye contact, attention, cause and effect relation and simple communication responses (Brodeur, 1987; Elefant & Lotan, 1998; Gage, 1987; Hadsell & Coleman, 1988; Wigram, 1991, 1995, 1997) and some expand their work with this population to a more psychodynamic orientated approach (Elefant & Lotan, 1998; Hill, 1997;
Montague, 1986; Wigram, 1995). Music can be used to promote emotional expression and act as a container for the person with Rett syndrome (Hill, 1997). It can help to reflect their mood change and support them when in different emotional states. Music can be used as an assessment tool or as a motivational factor that might enhance learning (Demeter, 2000).

Wigram (1995) attests to the emotional expression the girls reveal in their music. He suggests a more psychodynamic approach once the girl has become familiar and feels comfortable in the music therapy session with the therapist. He describes the girls attraction to music, but also their excitement to the interaction between the therapist in the musical games (Wigram, 1995). Wigram (in Burford and Trevarthen, 1997) is described interacting with a girl with Rett syndrome in a humorous and playful fashion. The musical jokes (in a form of surprising pauses) evoked the girl to laugh during these events. The relationship between the therapist and the girls is the basis of a successful intervention and building it gradually in a structural format is suggested. The stronger the relationship becomes, the more engaged the girls are and the sessions become with fewer boundaries and with more freedom (Hill, 1997; Wigram, 1991, 1995). It is important to work with their inner person and look for the smallest nuances (Hill, 1997; Montague, 1988), which can be achieved by a sensitive and finely, tuned therapist.

Wigram (1997) also conducted several in-depth assessment studies through vibroacoustic therapy in the treatment of girls with Rett syndrome. Vibroacoustic therapy is a treatment where pulsed, sinusoidal low frequency tones between 35-80 Hz. are combined with relaxing music, and played through bass speakers built into a treatment couch or chair. Measured on normal subjects in a clinically controlled trial, the effect is to reduce levels of energetic arousal, general arousal and tension arousal, as well as heart rate (over time). As the Rett population typically suffers from states of over-arousal, vibroacoustic therapy was considered a relevant intervention (Wigram, 1996; Wigram, 1997). The assessments undertaken by Wigram looked at changes that took place during vibroacoustic therapy and these
included changes or reductions in hand stereotypic behaviors, in hyperventilation, in muscle tone and arousal levels, improved relaxation and visual evidence of interest and pleasure. Anecdotal results showed that this treatment approach had positive effect on this population (Wigram, 1997). The girls relaxed while on the vibroacoustic couch, there was a reduction of hyperventilation, and decrease of hand stereotyped movements, reduction of movement and relaxation of muscle tone. Some girls vocalized during the therapy session or when the music was turned off as if using vocalization to communicate pleasure or dissatisfaction.

Clinical experiences have shown that despite severe motor disability in the population of Rett syndrome, music has been a very important motivating stimulus (Wigram, 1995). Through music they can express some of their feelings and needs. Sometimes those feelings reflect fun and enjoyment, while at other times they are more reflective, even melancholy and sad. Expressing different types of feelings enhances their motivation and the urge to learn. Since motivation is one of the most important key elements in learning, music therapy can provide a pathway to the development and learning in girls and women with Rett syndrome. Perhaps these channels Hill (1997) refers to may be discovered and opened by establishing opportunities for individuals with Rett syndrome in which they can demonstrate their ability to participate and to succeed. There are many opportunities to ‘succeed’ through music and it is evident that with each success there is a drive to attempt new challenges and to continue to grow. Music gives a direct gratification, so when a child chooses a song and the song is immediately sung the reward for the child with Rett syndrome is substantial.

Individuals with Rett syndrome have given definite indications as to their likes and dislikes of musical stimulus. It has been reported that girls like listening to music and some would tap along with music and dance by swaying back and forth. (Holdsworth, 1999; Lindberg, 1991; Merker et al., 2001). In one survey, most girls made spontaneous sounds and a few used simple words while hearing music (Lindberg, 1991). Individuals with Rett syndrome may not initially vocalize. Once
they have established a closer rapport with the music therapist, they may not be as inhibited during which there is evident to the development of vocalization and a beginning of vocal sounds. These intentional vocal sounds are initially produced very gently with soft resonance, while with time the child will develop a strong open sound. The sounds become varied and the child seems to have more control over her own vocalization. The development of vocalization can be seen as an extension of her development of self-confidence and follows along the developments in her play (Hill, 1997). The vocalization becomes interactive between the therapist and the child, a developed means of communication. Wigram & Cass (1996) describe one girl who during assessment vocalized to a familiar tune with much joy and once the music was turned off the vocalization changed to sounds reminiscent of an unpleasant reaction. The music was turned on once more, and the girl smiled and content vocal sounds were heard.

Burford & Trevarthen (1997) suggest to encourage and to strengthen this ability of vocal interaction, which will help to promote motivation and thus result in better communication. Once the child uses her vocalization as a form of communication, she gains the potential to reveal her more intimate inner feelings (Wigram, 1991). When this type of relationship has been established and the child has a place to express her emotions, the use of music can have a greater impact (Hill, 1997; Montague, 1988). Music can then be utilized to enhance cognitive development and to augment her communication abilities. Learning can take place once she has established a relationship with the therapist and has a secure place to express her emotions (Hill, 1997; Montague, 1988; Wigram, 1991; Wigram & Cass, 1996).

Teaching communication skills is extremely important for the Rett syndrome population, but these must have relevance to their daily environment. Since music is very meaningful and is a motivational force for them, using songs can be a ground base to enforce communication. Pre-composed songs as part of music therapy intervention have been used successfully with the Rett syndrome population (Brodeur, 1987; Hadsell & Coleman, 1988; Elefant & Lotan, 1998;
Hetzroni & Rubin, 1998; Wigram, 1991; Wigram & Cass, 1996; Wylie, 1996). They seem to respond very positively to songs, nursery rhymes and other tunes they may be familiar with (Wigram, 1991; Wigram & Cass, 1996), it's repeated rhythmic patterns and melodic phrases and are described as ones who “may exhibit sensitivity to playful teasing” (Burford & Trevarthen, 1997, p.3). Songs have been used to elicit positive social interaction and to develop eye contact in the Rett syndrome population (Hadsell & Coleman, 1988). The repetitions in the songs help build anticipatory response and these enhance the girls’ involvement in the musical interaction (Brodeur, 1987; Hadsell & Coleman, 1988).

Individuals with Rett syndrome are believed to have musical preferences and favorites (Holdsworth, 1999; Merker et al., 2001). In a survey by Holdsworth (1999) music is described as a means to a positive mood change if familiar, and negative reactions are sometimes observed to unfamiliar music. When unfamiliar music is introduced diminishing responsiveness is viewed when one compares levels of responsiveness to those found when familiar music is presented. They seem to prefer simple, bright and energetic music. Other cases have been reported of some girls who show preference for a particular song by smiling and becoming more animated as these familiar songs are sung (Elefant & Lotan, 1998; Hadsell & Coleman, 1988; Holdsworth, 1999; Woodyatt & Ozanne, 1992, 1994). Their recognition of favorite songs was also characterized by increasing activity levels and hyperventilation (Woodyatt & Ozanne, 1992, 1994). On the other hand, singing and slow music has been found to relax, but in some cases to increase anxiety levels in girls with Rett syndrome (Mount et al., 2001)

In a study performed by Hetzroni and Rubin (1998), familiar songs were successfully applied as one of the domains in a computer-assisted instruction study which investigated girls with Rett syndrome’s ability to identify ‘Augmentative and Alternative Picture Symbols’. Singing familiar songs has been an excellent motivator to get individuals with Rett syndrome to attempt one-handed tasks such as touching a selected picture (Elefant & Lotan, 1998; Hadsell & Coleman, 1988)
or as an accompaniment for promoting hand use with musical instruments or objects (Wylie, 1996).

In Beit Issie Shapiro in Israel, a center for children with developmental disabilities (seven girls with Rett syndrome are enrolled), some of the girls hum or vocalize to favorite songs. Some girls may respond or imitate vocalization made by the music therapist. Due to my own personal experience of the important role of music in the lives of individuals with developmental disabilities as a group and Rett syndrome in particular, and in light of the contradicting reports on communication and cognitive skills of Rett syndrome, my curiosity to see whether the clinical opinion of many different therapists and teachers could be found to be true in an experimental study. Although some reports suggest that individuals with Rett syndrome can only reach a pre-intentional level of development my experience has made me think differently. Using music (songs in particular) as a therapeutic tool enables the therapist to uplift the child with Rett syndrome to high levels of cooperation.

The primary objective of this study was to find out if it is possible to establish reliable communication skills through the use of songs in therapy. The research study set out to see if the girls could identify the songs and to make their choice of preferred song. Although most girls with Rett syndrome show a very impaired level of functioning, their desire to hear their chosen songs may enable them to use eye gaze, head nod, nose pointing, hand reaching, and any other bodily tool at their disposal to indicate choice.

The study also set out to gather examples of non-verbal communication symbols such as smiles, laughter, facial expression, and to try to process and translate it into a meaningful expressive communication.
The purpose of this research was therefore to investigate the following hypothesis. *Can songs in music therapy enhance communication in girls with Rett syndrome?* The sub questions supporting the primary research question were:

1. *Are girls with Rett syndrome able to make intentional choices?*
2. *Are girls with Rett syndrome able to learn and sustain learning over time?*
3. *Do girls with Rett syndrome reveal consistent preferences through choices they make?*
4. *How do girls with Rett syndrome demonstrate emotional and communicative behaviors?*
CHAPTER TWO

METHOD

2.1 DESIGN

A single case, multiple probe design was used to evaluate individual choice of, and response to, familiar and unfamiliar songs. This method of research is a form of multiple baseline design, which enables several comparisons of behaviors, responses, and musical elements to be made within each case. The experimental investigation is within treatment of the individual subject (Barlow & Hersen, 1984; Cooper, Heron, & Heward, 1987; Kazdin, 1982; Kratochwill, 1992).

The efficacy and value of single case design in quantitative and qualitative research has become increasingly recognized over recent years. Single case research design appears to have originated within quasi-experimental investigation and applied behavior analysis (ABA). It has developed and its usefulness has become apparent within a variety of professional fields, such as: clinical psychology, special education, social work and research on communication disorders (Kratochwill, 1992). It is one of a spectrum of case-study research methods applied to the investigation of individual change in clinical practice (Aldridge, 1996).

This method of research with built-in replication focuses on evaluation of the outcome of specific intervention techniques (Kratochwill, 1992; Sevcik, Romski, & Adamson, 1999). Repeated measurements are taken over an extended period of time.
in a single case study under precise and standardized conditions (Barlow & Hersen, 1984). In a ‘time series design’, measurement is taken at intervals during the course of the investigation. There are several approaches in single case design, some of which rely on establishing baseline measurements. In order to evaluate change over time, a number of new baselines may be taken in a ‘multiple baseline design’.

The research model chosen for this study was a multiple probe (an alternative to multiple baseline), a form of single case design, which is a highly flexible, enabling the analysis of the effects of the independent variable across multiple behaviors, settings and/or subjects (Cooper et al., 1987). Multiple-baseline design relies on the examination of performance across several different baselines in order to draw inferences or interpretations regarding the effect of treatment. The effects are demonstrated by introducing the intervention following baselines at different points in time (Kazdin, 1982). When a specific behavior has achieved a stable and pre-determined level of response under the treatment condition or reached a pre-established criterion, the independent variable is applied to the next behavior under investigation. If this second behavior changes in the same manner as behavior one, replication of the independent variable's effect has been achieved. After behavior two has reached the pre-determined level, the independent variable is then applied to behavior three. In order for the multiple baseline design to achieve rigor as a research method, it is recommended that it will be carried through several replications (Cooper et al., 1987; Kazdin, 1982). In this study trials evaluating behavior in the subjects were replicated.
In the initial period of observation, repeated measurements of the target behaviors under investigation are made. This initial period is defined as the ‘baseline’ (Barlow & Hersen, 1984; Cooper et al., 1987). Separate baselines are taken on selected behaviors in a particular case. A baseline establishes the current level of performance, and then becomes feasible to predict future performance or measure change. The purpose of the baseline measurement is to obtain a standard measure at a point in time of the efficacy of an intervention (Aldridge, 1996; Barlow & Hersen, 1984; Kazdin, 1982). It demonstrates the effect of the intervention by showing that behavior changes only when the intervention is applied (Kazdin, 1982). The baseline must demonstrate stability and consistency before the intervention is introduced. It is advisable to have a minimum of three separate observation points during the baseline to establish a trend in the data (Barlow & Hersen, 1984; Cooper et al., 1987). In this research study separate baseline measures were taken each time a new set of songs was to be introduced.

After a stable response level to picture symbols of songs had been confirmed by means of baseline measurements, the treatment was applied. The time needed to establish a stable baseline depended on the behavior of each participant, and ranged from four to five sessions.

The multiple probe design used in this research, a variant of multiple baseline design, is particularly appropriate when evaluating the effects of instruction on skill sequences (Barlow & Hersen, 1984; Cooper et al., 1987) and in testing for possible learning effects occurring to occur during treatment.
If baseline control is applied during the intervention period, there will be interruption of the intervention, and with the population under investigation here, an interruption can easily destabilize or disrupt carefully established learning processes. Therefore, periodical assessment (in the form of probes) is used that will enable researchers to evaluate learning process over time. Probes used as assessment for baseline or maintenance are measured throughout the intervention at regular intervals (Barlow & Hersen, 1984; Kazdin, 1982). The probe provides the basis for determining whether behavior change has occurred when compared with baseline measurements taken prior to intervention, or as an assessment a maintained skill or response (maintenance probe) for determining internalization of a learned behavior or response.

The data that emerges from such single case design research is typically analyzed by means of graphs that present results visually, or by descriptive statistics. In this study, ‘Effect Size’ measurements were applied in addition to graphic and descriptive statistics that describe change over time, to establish any effects resulting from intervention as compared with baseline levels of behaviors.

**Visual /Graphic analysis:**
Visual/graphics analysis is the most commonly used method of evaluating and presenting results within single-case research (Barlow & Hersen, 1984; Cooper et al., 1987). Visual analysis is a clear and informative means of determining the presence and consistency of experimental effect in analyses looking at behavior over time. The level, trend, and variability of the data must be evaluated within and across each phase of the experiment (Cooper et al., 1987). The graphs can describe a) available data and results, b) the process of the whole study, c) exact description of pre- and post-intervention and d) the duration of the study. In the graph, causal relationships between the intervention and behavior change can be seen (Barlow & Hersen, 1984).

**Effect Size analysis:**

In order to evaluate the effects of the intervention, an Effect Size was calculated in each case. ‘Effect Size’ is a way of quantifying the effectiveness of a particular intervention, relative to some comparison such as with a control group, or with baseline measurements in the case of pre-post test data. The routine use of Effect Sizes has often been limited to meta-analysis – for combining and comparing estimates from different studies – and is all too rare in original reports of educational research. Nevertheless, Effect Size is an important tool in reporting and interpreting effectiveness. The effect size is the standardized mean difference between the two groups. In other words:

$ES = \frac{\text{[Mean of experimental group] – [Mean of control group]}}{\text{Standard Deviation}}$
Effect size measures can be directly converted into statements about the overlap between the two samples in terms of a comparison of percentiles. For instance, an effect size of 0.8, means that the score of the average person in the experimental group exceeds the scores of 79% of the control group (Coe, 2000). Thus an Effect Size value of more than 0.8 is considered a highly significant effect representing the difference between treatment scores and control/baseline scores.

**Data collecting**

All sessions in the study were videotaped continuously throughout each session by two cameras. Both cameras were set in operation before the participant entered the therapy room to prevent their use influencing the girls’ behavior. The participant was seated at her chair and had a short acclimation period of 2 minutes after entering the room. At that time the researcher sat down, opposite the participant, conversing with her and responding to her mood (Hartman, 1984).

The independent variable or treatment variable in this study was comprised of 18 selected familiar and unfamiliar songs. These were sung accompanied with the guitar by the investigator, in response to the participant’s choice. The songs are described in the section on materials.

The dependent variables are behaviors presented by the participants during the course of all trials. As a result of previous knowledge, a survey of past articles on the topic of non-verbal communication (Sigafoos, 2000; Sigafoos et al., 1999, 2000) and careful preliminary observation of a few music therapy sessions, a set of behaviors was chosen to be observed and to be recorded. Those behaviors were:
- Intentional choices of songs by indicating picture symbols or orthographies that identified the songs.
- Response time in each trial
- Song preferences
- Affective responses (smiles, laughs, movements, etc.)
- Expressive vocalization
- Intentional speech
- Eye contact
- The presence or absence of stereotypical hand movements

Data collection forms were generated and prepared specifically for the study:

- **General record keeping form** – This form kept the trial dates, number of sessions, number of song sets, sequence in song sets, probes, maintenance and general comments (Appendix # II)

- **Direct/Video data collection form** – This form was used for direct and during later video observation. The form was marked during the trials by the researcher after each time the participant chose a song and confirmed her choice. At the end of each trial, the researcher kept a written log (on the same observation form) in which she described the participants’ emotional and communicative and pathological behaviors before, during and after the trial. (Appendix #III)

Further observation and confirmation of the data was through video observation, by the researcher and two other observers.
• **Behavior analysis form** - This form enables observation of behaviors during choice making activities (Appendix IV).

• **Communication, emotional and pathological observation form** - This form included behaviors when choosing the songs and during the singing of the songs. The data was collected during video observation (Appendix #V).

• **Duration of vocalization, emotional and pathological behavior form** – This form was constructed in the belief that fine differences could be detected in each participant in different situations, especially during vocalization. This form was not used eventually since no such differences were detected. (Appendix #VI).

• **Song selection frequency form** – This form was used to tally the amount of times each song was selected. (Appendix #VII)
2.2 PARTICIPANTS

Rett syndrome is rare in incidence and the recruitment and randomization of subjects to an experimental group relies on the availability of a large population from whom random selection can be made. This study employs single case design, and was undertaken at a center in Israel with an exceptionally large population of girls with Rett syndrome. Seven girls, ranging in age from 4 - 10 participated in this study (See table 1). All girls have been diagnosed with Rett syndrome according to guidelines established by the "Rett Syndrome Diagnostic Criteria Work Group" (1988, revised by IRSA 1997). Six girls were in stage III, the "plateau stage" of whom two are not ambulant. One girl was in stage IV, the "late motor deterioration stage". The girls' cognitive abilities were not evaluated, as standardized tools for measuring sequential and simultaneous processing of information, in order to determine intelligence levels in these girls as compared to other children of the same chronological age, cannot measure such severely handicapped individuals. Most test instruments, such as the WISC or the Kauffman ABC rely in part on verbal skills and good enough hand skills, both of which are significantly impaired or absent in this population.

The participants were enrolled at a center for special education in "Beit Issie Shapiro", a day center for developmentally impaired children in Raanana, Israel. Of the seven girls recruited for this study, four attended one class for children age’s seven to ten, while the three remaining attended another class for children age’s four to seven.
### Table 1 - Participant personal information

<table>
<thead>
<tr>
<th>Name</th>
<th>Age</th>
<th>Onset of regression</th>
<th>Ambulatory</th>
<th>Stage</th>
<th>Epilepsy</th>
<th>Verbal skills</th>
<th>Duration of pre-research MT</th>
</tr>
</thead>
<tbody>
<tr>
<td>Aviv</td>
<td>9.10</td>
<td>20 months</td>
<td>Yes</td>
<td>III</td>
<td>No</td>
<td>No</td>
<td>4.5 years</td>
</tr>
<tr>
<td>Elisheva</td>
<td>8.2</td>
<td>18 months</td>
<td>Yes</td>
<td>III</td>
<td>No</td>
<td>No</td>
<td>1 year</td>
</tr>
<tr>
<td>Hilla</td>
<td>10.2</td>
<td>24 months</td>
<td>Yes</td>
<td>III</td>
<td>No</td>
<td>Few words</td>
<td>4.5 years</td>
</tr>
<tr>
<td>Meirav</td>
<td>9.8</td>
<td>12 months</td>
<td>Yes</td>
<td>III</td>
<td>Yes</td>
<td>No</td>
<td>4.5 years</td>
</tr>
<tr>
<td>Rachel</td>
<td>6.5</td>
<td>18 months</td>
<td>No</td>
<td>III</td>
<td>No</td>
<td>No</td>
<td>1 year</td>
</tr>
<tr>
<td>Tali</td>
<td>4.8</td>
<td>12 months</td>
<td>No</td>
<td>III</td>
<td>No</td>
<td>No</td>
<td>1 year</td>
</tr>
<tr>
<td>Talia</td>
<td>10.7</td>
<td>18 months</td>
<td>No</td>
<td>IV</td>
<td>Yes</td>
<td>No</td>
<td>4.5 years</td>
</tr>
</tbody>
</table>

All seven girls had already been receiving music therapy for months and in some cases for years before the study began. A natural break (holiday period) occurred before the trials in this study started, during which no interventions were given. Four girls had individual and group music therapy with the investigator (a music therapist) until the beginning of the holiday break before the study commenced. Although the three other girls participated in individual and group music therapy with another music therapist, the investigator was a familiar figure to them as they had contact with her on many occasions in the center in situations other than music therapy. The individual and group music therapy sessions consisted of improvised music playing and singing. Some of the aims during the individual music therapy sessions were as follows: a) to allow the girls to express themselves emotionally by
actively playing instruments or listening to live music, b) to communicate through making their own choices as to what materials (such as: instruments or songs) they may want to use, c) to enhance functional hand use through playing different instruments, d) to provide a relaxed and calming atmosphere when the girls are agitated; and more. The individual music therapy sessions used improvised music more than pre-composed music and the therapist used mostly the piano and the guitar as well as vocalization.

The group music therapy was more structured and singing songs was one of the methods used very frequently during these sessions. Some of the goals for singing children’s song were: a) to enhance song repertoire for the children, b) to build independence - the children would indicate their song choice by looking, facial pointing, head nodding or touching with their hand the picture symbol, thus encouraging initiative, c) to elicit emotional and communicative responses during songs. The girls would typically express themselves emotionally (i.e. smile, laugh, vocalize, frown) and communicatively (i.e. by body movement, leaning, head turning, eye closure).

The girls in this study were familiar with song singing intervention, and also with some of the songs used in the trials. During the course of individual and group music therapy, all girls had been exposed to different levels of choice making activities. During group music therapy sessions, many of the activities had already involved the use of picture symbols to choose songs or instruments. Therefore the participants in the research were not naive or new to the stimulus, and this needs to
be taken into consideration when discussing both their response to the intervention, and their lack of response at baseline.

2.3 SETTING AND MATERIAL

The study was conducted in the music therapy room at the special education center "Beit Issie Shapiro". Myself, the center’s music therapist, conducted the research trials.

Two chairs were placed in the room facing each other half a meter apart. The girl (participant) sat on one chair while the music therapist (researcher) sat on the other, facing each other.

A set of cards containing Picture Communication Symbols (PCS) (Mayer-Johnson, 1981) was prepared on a 10X10- cm card. The set of cards contained reference to the songs with the title words written above the graphic picture symbol. A second set of card contained only orthographies (Hebrew words) for the song title, and these were displayed on 10 X 5 cm cards. A small piece of Velcro tape was attached to the back of each card so that it could be easily secured to and removed from a 50X30- cm communication board (made of a piece of tapestry). Hebrew is read right to left so the order of presentation of the visual symbols on the communication board was always right to left.

The researcher used a guitar to accompany her singing, and the typical style of accompaniment was chord strumming.

Two video cameras on tripods were placed in the room. One camera faced the participant and the other faced the investigator. There were a total of 18 different songs in the study. Some of them were familiar and some were unfamiliar to the
participants. The songs were divided into three ‘sets’, a total of six songs in each set (four familiar and two unfamiliar songs in a set). The songs were either traditional children songs, or songs the investigator had written. All songs had been translated into Hebrew. The content of the songs was about a number of subjects, including animals, body-parts, food items and transportation. All the songs were short with a very simple repeated verse format with contrasts in phrases built into the style of the songs. In some songs verses were repeated in different styles while others had one verse with last line repeating 2-4 times.

Table 2 – Song list with short description
The score of the songs can be seen in Appendix VIII.

Detailed analysis of the songs was planned following the trials in order to determine elements that evoked responses, and to gain an overview of style that determined preferred choice. Results and discussion of this qualitative analysis and inferences that can be drawn will be reported in Chapter 4.

<table>
<thead>
<tr>
<th>Song</th>
<th>Tempo</th>
<th>Key</th>
<th>Meter</th>
<th>Familiar/Unfamiliar</th>
<th>Short Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nut</td>
<td>144</td>
<td>A</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, jazzy, syncopated, begins with upbeat, vocal play/sounds</td>
</tr>
<tr>
<td>Monkey</td>
<td>184</td>
<td>D</td>
<td>2/4</td>
<td>Familiar</td>
<td>One verse, swing, begins with upbeat</td>
</tr>
<tr>
<td>Spider</td>
<td>104</td>
<td>A</td>
<td>6/8</td>
<td>Familiar</td>
<td>One verse, syncopated, vocal play</td>
</tr>
<tr>
<td>Bee</td>
<td>160</td>
<td>D</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, begins with upbeat &amp; fermata, vocal play</td>
</tr>
<tr>
<td>Train</td>
<td>132</td>
<td>A</td>
<td>6/8</td>
<td>Unfamiliar</td>
<td>One verse, begins with upbeat, syncopated, vocal play/sounds</td>
</tr>
<tr>
<td>Farm</td>
<td>132</td>
<td>C</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, vocal sounds</td>
</tr>
<tr>
<td>Bird</td>
<td>152</td>
<td>D</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, begins with upbeat, lightness, vocal sounds</td>
</tr>
<tr>
<td>Chick</td>
<td>120</td>
<td>D</td>
<td>3/4</td>
<td>Unfamiliar</td>
<td>One verse, rocking motion, begins with upbeat</td>
</tr>
<tr>
<td>Butterfly</td>
<td>100</td>
<td>A</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, begins with upbeat, lightness</td>
</tr>
<tr>
<td>Frog</td>
<td>112</td>
<td>A</td>
<td>2/4</td>
<td>Familiar</td>
<td>One verse, begins with upbeat, vocal sounds</td>
</tr>
<tr>
<td>Star</td>
<td>96</td>
<td>D</td>
<td>4/4</td>
<td>Unfamiliar</td>
<td>One verse, begins with upbeat, rocking motion</td>
</tr>
<tr>
<td>Tree</td>
<td>84</td>
<td>A</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, begins with upbeat, rocking motion, vocal sounds</td>
</tr>
<tr>
<td>Turtle</td>
<td>84</td>
<td>A</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, begins with upbeat, rocking motion</td>
</tr>
<tr>
<td>Hands</td>
<td>72</td>
<td>D</td>
<td>4/4</td>
<td>Unfamiliar</td>
<td>One verse, lightness</td>
</tr>
<tr>
<td>Crocodile</td>
<td>72</td>
<td>Chant</td>
<td>4/4</td>
<td>Unfamiliar</td>
<td>One verse, begins with upbeat, syncopated</td>
</tr>
<tr>
<td>Fish</td>
<td>92</td>
<td>A</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, rocking motion, one vocal sound</td>
</tr>
<tr>
<td>Elephant</td>
<td>90</td>
<td>A</td>
<td>4/4</td>
<td>Familiar</td>
<td>One verse, heavy motion</td>
</tr>
<tr>
<td>Rabbit</td>
<td>92</td>
<td>A</td>
<td>4/4</td>
<td>Unfamiliar</td>
<td>One verse, begins with upbeat, accented</td>
</tr>
</tbody>
</table>
2.4 PROCEDURE

Seven girls with Rett syndrome participated in individual research trials. The trials incorporated choice making with familiar and unfamiliar children's songs. During intervention trials, once the girls had made their choice, the songs were sung with guitar accompaniment by the investigator (a music therapist). The sessions were held three mornings per week and each lasted between 20 - 30 minutes. The duration of the study was five months and included baseline, intervention and maintenance trials. During the following three months an additional three maintenance trials were administered.

The participants chose one symbol out of groups of either two or four presented symbols. Whether they were presented with a choice of two or four was determined according to each girl's choice-making ability. Consistency and reliability of choice making ability was established in pre-baseline tests undertaken in preparation for the main study. Five girls were found to be able to choose effectively from a choice of two stimuli, while two girls demonstrated the ability to choose from a selection of four stimuli on presentation. It was decided to use four symbols or orthographies with these two more able girls (Aviv and Meirav), who could choose rather easily from a choice of only two symbols, whereas they had to develop more discrimination to choose from a choice of four. Development of choice making in these two brighter girls would demonstrate more complex learning ability by the presentation of more complex stimuli.
Six girls chose from picture symbol cards with song title on it, while one of the two more able girls (Aviv) revealed the potential to choose from orthography cards, on which were written the titles of the songs.

The picture symbols (representing the songs) were attached to a communication board with Velcro and were spaced approximately 5 cm apart, and presented about 30 cm in front of the girl’s face.

Before the music therapy intervention was applied, a stable baseline (with no music) had to be established, its purpose being to determine whether the participants could choose or not, and more specifically whether there was any consistency or reliability in their choosing that would confirm intentional choice.

The investigator asked each participant to indicate a song (represented by picture symbols) out of two (five girls) or four (two girls) possible choices. This choice indication was made through movement of the girl's hand, nose, head or eye gaze, according to her preference and ability. Once the preferred song was chosen, the order of the symbols was randomly changed out of sight. The communication board was then re-presented with the symbols in different positions, and the girl was asked to choose again to confirm her original choice. The purpose of this was to verify that her choice was intentional and specific. Immediately following her confirmation, the song was sung to her.
**Baseline procedure:**

The purpose of the baseline measurement was to determine the current level of the participants’ performance and to measure abilities to choose at a point in time that could later be compared with the intervention, in order to determine the efficacy of the intervention.

The baseline in the study consisted of two stages:

**Stage I:** During the baseline phase the participant was given opportunity to choose a preferred song. If she did not reply within 15 seconds, she would be given another opportunity to choose. The following procedure was practiced during baseline:

1) The communication board was empty of symbols and was presented facing the participant. The girl was requested to choose a song. The same wording of the request was used each time: "What song would you like to choose?" At the end of the question the symbols were revealed to the girl and attached one by one to the communication board while the name of each song was said out loud e.g. "monkey song, train song, bee song or nut song?"

If the girl did not reply within 15 seconds, she was given another opportunity to choose.

2) The same wording as the first time was used for the request. This time all symbols were already placed on the communication board and the investigator pointed to one picture at a time from top right to top left, then bottom right to bottom left (as the Hebrew language is read), naming each song.
If the girl did not reply within 15 seconds the next group of songs was introduced and it was checked that she had failed to choose.

**Stage II:** If the participant indicated a song by choosing (during the first or the second opportunity in part 1), her choice of song was taken off the communication board, and shown to her while the investigator said: "You chose the song about the (e.g.) monkey".

The communication board was then turned around (towards the investigator) and the position of the symbols on the board was randomly changed. Then the board was turned back towards the girl, and she was asked to show her choice one more time in these words: "Can you please show me the song about e.g. monkey one more time, but I will not sing the song to you this time". The purpose of this was to verify that her choice was intentional and specific for a preferred song.

If she confirmed the song within the span of 15 seconds, the next group of songs within the same set was visually presented. If she did not confirm the song she had chosen on the first occasion, a second opportunity to choose was given to her. This time the therapist said: "You chose the song about the e.g. monkey (pointing at the monkey picture). Can you please show me the song about e.g. monkey one more time (no pointing), but I will not sing the song to you this time."
**Intervention procedure:**

The intervention phase is when the musical stimulus (the songs) was introduced. The intervention contained two parts.

**Stage I:** When the therapist asked the girl to choose a song.

**Stage II:** After the girl had chosen a song, the position of the symbols on the board was again changed out of sight of the girl, and she was then asked to confirm her choice one more time. She was told that if she confirmed her choice, the song would be sung to her. The intervention had a structural hierarchy of prompting. Each part contained five steps in a hierarchy progression. The steps were graded from the most independent choice making (with no physical cues or assistance) to the least independent, where physical prompting and cues were utilized. Only the first two steps (1-2) were counted as independent choice making and were included in the final data results. The three other steps (3-5) had prompting and could not be viewed as independent choice making. If a participant’s choice making went into steps 3-5, this data was not eligible for inclusion as a confirmed choice.

**Figure 1** illustrates through a flow chart how the various steps in the choice-making procedure were followed through, and decisions were taken at each point in the process.
**Figure 1 - Song selection flow chart**

**Stage I selection**

1. **Song presentation (Step 1)**
   - 15 seconds
   - No choice
   - Choice

2. **Song presentation (Step 2)**
   - 15 seconds
   - No choice
   - Choice

3. **Song presentation (Step 3)**
   - 15 seconds
   - No choice
   - Choice

4. **Song presentation (Step 4)**
   - 15 seconds
   - No choice
   - Choice

5. **Song presentation (Step 5)**
   - 15 seconds
   - No choice
   - Choice

Starting a new trial

**Stage II – confirmation**

1. **Song presentation (Step 1)**
   - 15 seconds
   - No choice
   - Choice

2. **Song presentation (Step 2)**
   - 15 seconds
   - No choice
   - Choice

3. **Song presentation (Step 3)**
   - 15 seconds
   - No choice
   - Choice

4. **Song presentation (Step 4)**
   - 15 seconds
   - No choice
   - Choice

5. **Song presentation (Step 5)**
   - 15 seconds
   - No choice
   - Choice

Singing the selected song
The procedure demonstrated in Figure 1 is complex and requires a clear explanation as to how the research process was carefully followed in all trials.

**STAGE I (Indicating song choice)**

**Step 1** - Just as in the baseline phase, the communication board was empty and faced the girl. The participant was requested to choose a song. The same wording of the request was used each time: "What song would you like to choose?" At the end of the question the symbols were exposed and attached one by one to the communication board while the names of each song was announced e.g. "monkey song, train song, bee song or nut song?"

If the participant indicated her choice within 15 seconds, she was introduced to step 1 in Stage II of the intervention. If the girl did not indicate her choice within 15 seconds, step 2 of Stage I was followed.

**Step 2** - The same verbal procedure was used for the request that the girl choose a song as in step 1, only this time all symbols were on the communication board (left from step 1). The investigator pointed to one picture at a time from top right then top left (if choosing between two songs) & bottom right then bottom left (if choosing between four symbols just as the Hebrew language is read), naming each song.

If the girl indicated her choice within 15 seconds, she was introduced to step 1) in Stage 2 of the intervention. If the girl did not indicate her choice within 15 seconds the third step was introduced and was considered as prompting.
**Step 3** - The same verbal instructions was used for the request that the participant choose a song as in step 2, only this time when naming the songs, the researcher used ‘hand over hand’ assistance to prompt her to finger point at the symbol. Her finger was then freed to give her an opportunity to choose independently.

If the girl indicated her choice within 15 seconds, she was introduced to step 1 in Stage II of the intervention. However successful choosing during step 3 was not included in the data analysis. If the girl did not indicate her choice within 15 seconds, the fourth step was introduced.

**Step 4** - The same verbal cues was used for the request that the girl chooses a song as in step 2, with hand over hand prompting (as used in step 3). In addition the investigator named the songs followed by singing the beginning phrase of each song, During that time the girls' finger was pointing at the symbol with hand over hand prompting. The girl's finger was then freed to give her opportunity to choose independently.

If the girl indicated her choice within 15 seconds, she was introduced to step 1 in Stage II of the intervention. If the girl did not indicate her choice within 15 seconds, the fifth step was introduced.

**Step 5** - All symbols were taken off the communication board. The girl was asked to choose a song as in previous stages. The symbol was attached to the board while naming it. The investigator then sang the whole song. The next song was introduced
and then sung. The girl was then asked to choose a song using the same verbal procedure as in step 2.

If the girl indicated her choice within 15 seconds, she was introduced to step 1 in Stage II of the intervention. If the girl did not indicate her choice within 15 seconds, the next group of songs was introduced.

**STAGE II (confirming song choice)**

Once the preferred song was chosen, the order of the symbols were randomly changed on the communication board, out of site of the participant, and then re-presented to her. On re-presentation, the participant was cued verbally to indicate her choice of her preferred song, thus confirming her previous choice. The purpose of this was to verify that her choice was intentional. Immediately following her confirmation of choice, the song was sung to her.

Stage II also contained five hierarchical steps from the most independent (steps 1-2) to the least independent and with most prompting (steps 3-5). If the girl indicated her choice during Stage I step1, she would move onto Stage II step 1. Only when a clear choice had been made during the first two steps were these choices counted in the final results, as this indicated independent and intentional choice making.

**Step1** - Just as in the baseline, the chosen song was taken off the communication board, shown to the participant while the investigator said: "You chose the song about the e.g. monkey". The communication board was then turned around (towards the investigator) and the position of the symbols on the board was randomly changed
out of sight of the participant. While the board was turned back towards the participant, she was asked with the following verbal instructions to show her choice one more time: "Can you please show me the song about e.g. monkey one more time and I will then sing you the song?" The song was sung immediately after the girl confirmed her choice. The purpose was to verify the intention of her choice. If the girl did not confirm her choice within 15 seconds, the second step was introduced.

**Step 2** - The same wording as the first step was used only this time the investigator pointed at the chosen song and saying: "You chose the song about the e.g. monkey. Can you please show me the song one more time and I will then sing you the song?"

If the participant did not confirm her choice within 15 seconds the third step was introduced and was considered as prompting.

**Step 3** - The exact same procedure as the second step, only this time when naming the chosen song, the girl's finger pointed at the symbol with hand over hand prompting. The girl's finger was then freed to give her opportunity to choose independently.

The song was sung immediately after she confirmed her choice (within the 15 seconds). The purpose was to verify the intention of her choice. If the participant did not confirm her choice within 15 seconds, the fourth step was introduced.

**Step 4** - The exact same procedure was applied as in the previous step. The investigator named the chosen song followed by singing a phrase of it while the
girl’s finger was pointed at the symbol with hand over hand prompting. The girl's finger was then freed to give her opportunity to choose independently.

The song was sung immediately after the girl confirmed her choice (within the 15 seconds). The purpose was to verify the intention of her choice. If the girl did not confirm her choice within 15 seconds, the fifth stage was introduced.

**Step 5** - All symbols were taken off the communication board and the chosen song was attached to the communication board. The therapist indicated that the girl had chosen the particular song and then she sang the entire song to the girl.

Step 1 in Stage I of the next group of songs was introduced immediately following each successful confirmation of choice or at the end of step 5 in stage II. As previously mentioned, there were a total of 18 children’s songs, both familiar and unfamiliar used in this study. The songs were randomly divided into three sets with a total of six songs in each set (four familiar and two unfamiliar songs in a set).

The method by which these songs were presented is illustrated in an example form in Table 3.

**Table 3 - Example of choice options in a set of six songs**

<table>
<thead>
<tr>
<th>Song groups</th>
<th>Trial 1</th>
<th>Trial 2</th>
<th>Trial 3</th>
<th>Trial 4</th>
<th>Trial 5</th>
<th>Trial 6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Choice symbols</td>
<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>from 2</td>
<td>A B</td>
<td>C D</td>
<td>E F</td>
<td>A C</td>
<td>D F</td>
<td>B E</td>
</tr>
<tr>
<td>Choice symbols</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>from 4</td>
<td>A B</td>
<td>E B</td>
<td>C E</td>
<td>F A</td>
<td>B F</td>
<td>A C</td>
</tr>
<tr>
<td></td>
<td>C D</td>
<td>A F</td>
<td>F D</td>
<td>E D</td>
<td>C D</td>
<td>B E</td>
</tr>
</tbody>
</table>
The six songs in each set were divided into groups. For the five participants who chose from only two songs, the six songs in each set were paired into six different groups of two songs. For the two more able subjects who chose from four songs, the six songs in the set were randomly grouped into six different groups of four songs. Every trial (during Stage I and Stage II procedure), a group of two or four symbols were randomly chosen. Five of the participants, who chose out of two symbols of songs, had the opportunity to see each song twice in the set presented. The two participants that chose between four symbols had the opportunity to see each song four times in the set presented. Each participant had a total of six trials each session when she could choose and hear up to six songs. She only heard the song when she had confirmed her choice, completing Stage I and Stage II of the procedure, as defined above.

The criterion for moving onto the second and then third set of songs was pre-defined. It was determined so that the length of time will show stability in the intervention. The participant moved onto the next set of songs after she had successfully chosen songs and confirmed her choice of a specific song in five out of six trials in total. These choices had to be made in three out of four sessions to demonstrate consistency and reliability.

**Probe:** Baseline measurement was established with each girl (typically 4-5 sessions) before the intervention was applied. All three sets of songs were presented in one session for the baseline. Once the intervention had been introduced, starting with Set 1 songs. During the treatment, ‘probe measurement’ was taken every fourth session. This continued during the second and third sets of songs as they were presented.
These were measured as well as when the next set was introduced. The probe technique continued not only as measuring baseline, but also as measuring maintenance of skills with sets that were completed.

**Table 4 - Research design, a schematic display**

<table>
<thead>
<tr>
<th>Set 1</th>
<th>Set 2</th>
<th>Set 3</th>
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</thead>
<tbody>
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</table>

<table>
<thead>
<tr>
<th>Session cycle</th>
<th>1</th>
<th>2</th>
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<th>7</th>
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<tbody>
<tr>
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<td>Baseline</td>
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<tr>
<td>Intervention set 1</td>
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<td>Intervention set 3</td>
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<tr>
<td>Baseline probe</td>
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<td>Maintenance probe</td>
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During the sessions when probe measurements were taken, the length of the sessions became longer than when the intervention consisted of one set. The intervention ended when each girl had fulfilled the criterion with the completion of all three sets of songs. The investigator did not know the duration time of the intervention, as each participant was to complete all three sets at her own individual pace. When the intervention was completed the maintenance procedure was applied.

**The Follow-up Maintenance Probe Sessions (FMPS):**

The FMPS was applied three times, during the weeks after the trials had been completed once the criterion of all three sets was achieved. It was determined before the study commenced. The first FMPS were implemented two weeks after termination of the intervention trials. The second FMPS four weeks after the first
maintenance trial (six weeks after termination of the intervention trials) and the third FMPS was implemented six weeks after the second maintenance trial (twelve weeks after termination of the intervention trials). Therefore total time between the end of the intervention phase and the final completion of the study was three months.

All session’s baseline, intervention, probe and maintenance were recorded by two video cameras. During the sessions data was recorded onto observational sheets. The video material was viewed and data was collected and transferred into observational sheets as well. The data was analyzed through the tools described above and the results are presented in the following chapter.

**Validity**

Validity has not received much attention in observational research and has been considered “inherently valid insofar as they are based on direct sampling of behavior and they require minimal inference on the part of observers” (Hartman, 1984, p. 129). Although each of the traditional types of validity is relevant to observation systems, content validity is especially important in the initial development of behavior coding system (Hartman, 1984), as is the case of the present research. All variables assumed to be relevant to indicate behaviors were gathered and categorized. The fields and categories designated in the forms used during the research were evaluated by the researcher, and by two other experts in the area of communication, mental retardation, and Rett syndrome, to ensure content validity

**Reliability**
The present study was aimed at observing and measuring different behaviors as indicators of intentional choice making and communicational abilities during music therapy sessions. Since observational instruments require periodic assessments to ensure intervention effectiveness (Hartman, 1984) and in order to collect the data needed in a reliable fashion, records (videotaping) were collected continuously.

The videotapes were observed and analyzed by three different observers at different times, using the video recording form as a mutual reference point, thus controlling possible biases (Hartman, 1984).

Intra-Observer Reliability
In order to establish intra-rater reliability the researcher (one of the observers) observed the video materials and her results were compared with her notes taken at the time of the trial sessions.

Inter-Observer Reliability
Both the researcher and two independent evaluators measured choice making. Inter-observer reliability was included in the design. Samples of video recorded trials were randomly selected and both independent observers separately scored results on prepared forms to evaluate intentional choice making. This process was carried out after, and separately from the researchers own scoring of video material. Agreement between observers of intentional choice making was established using a randomly selected 20% of the data.
The behavior and the cases in the study were repeated for subjects during the trials, with consistent procedures and responses, indicating that the study can be replicated with good expectations of reliability in the method, while nevertheless still allowing for individual differences in presentations of songs.
CHAPTER THREE

RESULTS

INTRODUCTION

The result chapter will address the questions derived from the main hypothesis in the order that they were presented. These will be analyzed according to the following topics:

- Intentional choice making
- Response time
- Learning process
- Song preference
- Emotional, communicative and pathological behaviors

Due to the nature of this research design (a multiple probe, single case design), the data collected will be processed for each participant separately (in alphabetical order). Thereafter, all cases as a group will be presented, with the intention of drawing out some generalizations that may be considered applicable to the wider population of females with Rett syndrome.
Reliability

All material in the study was video taped and collected into observational data sheets. All data was viewed for intra-observer reliability both for dependent and independent reliability and resulted in 100% accuracy. To ensure inter-observer reliability, 20% of the data was randomly selected, observed, and scored by an independent observer (a music therapist) which, found 96% observer agreement with the researcher in the dependent variable reliability and 99% observer agreement in the independent variable reliability. The independent reliability data measured the implementation of the researcher’s procedures in the following areas: (a) providing correct stimulus (picture symbols), (b) providing correct verbal cues, (c) waiting 15 seconds between trials, and (d) keeping the presentations of the songs consistent in length.
3.1 INTENTIONAL CHOICE - DATA ANALYSIS

Introduction

Intentional choice making is understood to be a process where a person is able to repeat and therefore to confirm an initial choice made by that person.

In order to detect the ability of intentional choice making in the population with Rett syndrome, each child was presented with a choice of orthographies or pictures symbolizing songs (either two or four depending on their level of ability) and asked to choose one. This procedure is further explained in detail in the method section (Figure 1, pp.97)

Thus, in every trial during baseline, intervention and maintenance, each participant was requested to make her choice from the orthographic or picture symbols presented, and then to repeat her choice for the purpose of confirming that she was choosing consistently and intentionally. The results were formulated into graphs to visually describe the data. The numerical scores for each girl in set 1, set 2, and set 3 songs comparing baseline with intervention, and baseline with maintenance were summed into means with standard deviations, and effect size calculations were made.

The graphs are to be read as follows:
In each session (axis x), the participant could choose and hear up to six songs (as indicated in axis y). The number of times the participant chose in each session was counted and indicated on the graphs. The blue diamond (at the top of the graph) represents the number of songs the girl chose the first time she was asked to choose. The red squares (at the bottom of the graph) represents when she confirmed (repeated) her choice. The confirmation of the choice (the red squares) was taken as a verification of the choice for this study, and provided the evidence of intentional choice making. The vertical lines (at the center of the graph) separate the baseline
trials from the intervention trials. The vertical dotted line separates the intervention trials from the maintenance trials.

Baseline for all sets (all 18 songs) was presented simultaneously during the same trial and is documented in the graphs (Figures 2-8). Four or five trials were undertaken at baseline to establish what the typical response of each subject was when the musical stimulus was NOT presented. After these 4-5 trials, the baseline period was ended and intervention for set 1 began. Baseline probes for sets 2 and 3 were continuously taken at equal intervals. Once the criterion for set 1 was achieved (three consecutive trials where following choice of songs, confirmation was made a minimum of 5/6 times), intervention for set 2 began. The researcher still continued to take maintenance probes at equal intervals for each following set (see procedure section in the method chapter, Table 4 research design schematic display, pp. 104).

The baseline and maintenance probes are illustrated in Figures 2-8 when a symbol of a diamond (first choice) and/or a square (confirmation of choice) stand alone and unconnected with other diamonds and squares.

This study comprises seven individual cases, and the results are presented correspondingly. Relevant interpretations and explanations of these results are included within each case in the result section. General inferences relating to these results both for individual girls and for the cohort as a whole will be addressed in the discussion.

3.1.1 Intentional choice in individual cases

The following data is presented in graphs, percentages and effect sizes to describe the consistency of intentional choice making. The graphs show baseline, intervention and
maintenance scores in all seven girls, and the raw scores from the graphs are then converted into percentages.

**Effect Size Calculations:**
In order to determine effect size calculations at a reliable level, the scores from all three sets presented in baseline, intervention and maintenance conditions were summed, averaged and the standard deviations were calculated. The effect of both intervention and maintenance were then calculated by subtracting the mean score of the baseline from the mean score of the intervention (or maintenance), and dividing it by the standard deviation from the baseline scores.
AVIV

Baseline:
During baseline set 1, Aviv responded almost always the first time she was asked to choose and chose 28 songs out of 30 possibilities (93%). During set 2, 24/30 possible songs were chosen (80%) and during set 3, 20/30 songs were chosen (66%). When asked to confirm her choices (baseline sets 1, 2 and 3), Aviv showed very low rates of intentional choice making: 5/28, 5/24 and 3/20 respectively (ranging between 20%-15%).

In Aviv’s case there seemed to be a gradual and consistent reduction of choice making between sets 1, 2 and 3. The reason for the gradual reduction in responding to the stimulus (orthographies) may be due to the fact that she realized that no songs would be sung after she had chosen. Her understanding that she will not be hearing song she had chosen, had led to a negative/rejecting reaction. One part of this behavior can be shown in the graph – a refusal to confirm her first choice. Another part of her reaction, where on several occasions Aviv turned away from the researcher or left her seat when asked to reconfirm her choice, cannot be shown in a graph.

Intervention:
The difference between the baseline and the intervention was evident. During the first three sessions in intervention set 1, there was a gap between the first time she made a choice and the confirmation of that choice. When she was first asked to choose, she responded almost a 100% of the time, but she only confirmed her choices consistently from session four. During the first three sessions there was less intentional choice making than thereafter. During intervention set 2 session 1, she demonstrated fewer (4/6) confirmations when compared with scores in the last three sessions of set 1, but became stable with higher (6/6) scores from session 2 and thereafter.
The reason for a large gap between making a choice in comparison to confirming the choice during the first three sessions of intervention set 1 could be due to the time it took Aviv to realize the difference between the baseline and the intervention sessions. From that point on, she responded almost 100% of the time and confirmed her choice consistently. The first part of the intervention period could be regarded as a learning period.

**Baseline probes:**

The baseline probes in sets 2 and 3 showed that most of the time Aviv did not confirm her choices, suggesting a strong lack of intentional choice making.

**Maintenance probes:**

During maintenance probes Aviv’s choosing was consistent during all choices presented both when first asked to choose and when asked to confirm her choice. During set 1 she confirmed her choice 16/18 songs (89%) and in set 2, 5/6 songs (83%).

**Maintenance:**

During maintenance sessions Aviv chose and confirmed her choice 18/18 songs in all three sets (100%).
Effect size calculation of Intervention and Maintenance compared with Baseline on the scores of all three sets presented

Baseline to intervention:
Baseline Pre-test scores = 2 0 1 1 3 0 1 1 2 0 1 0 0
Intervention Post-test scores = 3 2 3 5 5 5 4 5 6 5 6 6 6
Mean pre-test scores = 0.93. Mean post-test scores = 4.69
Standard deviation of pre-test scores = 0.88
Effect size = (4.69 - 0.93) / 0.88 = 4.27
**Baseline to maintenance:**

Baseline Pre-test scores = 2 0 1 1 1 3 0 1 1 1 2 0 1 0 0

Maintenance Post-test scores = 5 5 5 6 6 6 6 6 6

Mean pre-test scores = 0.93. Mean post-test scores = 5.67

Standard deviation of pre-test scores = 0.88

Effect size = \( \frac{5.67 - 0.93}{0.88} = 5.39 \)

The scores from both the intervention phase, and the maintenance phase when compared with the baseline scores show a very large effect, with an increased effect noted in the maintenance scores over baseline when compared with intervention.

**ELISHEVA**

**Baseline:**

Choice making in all three sets during baseline was inconsistent, showing drastic changes between sessions in each set. When asked to choose a song, Elisheva indicated 8 out of 30 songs, 13/30 songs, and 11/30 songs (in sets 1, 2 and 3 respectively). When confirming her choice, she was quite inconsistent, and low rates of intentional choosing were apparent: 3/8 songs, 5/13 songs, and 3/11 songs (in sets 1, 2 and 3 respectively)

**Intervention:**

There was a change once the intervention began. Elisheva chose 25/30 songs (83%), 17/18 songs (94%), and 23/24 songs (96%) in sets 1,2 and 3 respectively. A clear change occurred during intervention in set 1, session 3. From that point on Elisheva kept a choosing rate close to 100% for the rest of the intervention sessions. Intentional choice making was evident at this point and stood at a high rate of 19/25 songs (76%), 16/17 songs (94%), and 21/23 songs (91%) in sets 1, 2 and 3 respectively.
A steady and stable improvement was seen from the moment Elisheva was introduced to the music. This short period (three sessions) probably represents Elisheva’s learning period and the time it took her to understand that “now” she would hear the song she had chosen.

**Baseline probes:**
The baseline probes in sets 2 and 3 showed inconsistency in intentional choice making. Most of the time a low degree of confirmation of the choice was observed, thus a low intentional choice was evident during the baseline probes.

**Maintenance probes:**
Consistency was sustained during maintenance probes in set 1, which resulted in 18/18 songs (100%) and set 2 at 5/6 songs (83%).

**Maintenance:**
During maintenance sessions the intentional choice making resulted in 22/24 songs (92%), 24/24 songs (100%) and 17/17 songs (100%) in sets 1, 2 and 3 respectively.
Figure 3. Elisheva – selection and confirmation of songs.

Baseline | Intervention | Maintenance

Set 3
- # of songs selected in one session

Set 2
- # of confirmed songs in one session
**Effect size calculation of Intervention and Maintenance compared with Baseline**

**Baseline to intervention:**

Baseline Pre-test scores = 0 1 2 0 0 2 0 1 0 2 2 0 0 1 0
Intervention Post-test scores = 2 2 5 5 5 5 6 5 4 6 6
Mean pre-test scores = 0.73. Mean post-test scores = 4.67
Standard deviation of pre-test scores = 0.88
Effect size = (4.67 - 0.73) / 0.88 = 4.48

**Baseline to maintenance:**

Baseline Pre-test scores = 0 1 2 0 0 2 0 1 0 2 2 0 0 1 0
Maintenance Post-test scores = 6 5 6 5 6 6 6 5 6 6
Mean pre-test scores = 0.73. Mean post-test scores = 5.73
Standard deviation of pre-test scores = 0.88
Effect size = (5.73 - 0.73) / 0.88 = 5.68

The scores from both the intervention phase, and the maintenance phase when compared with the baseline scores show a very large effect, with an increased effect noted in the maintenance scores over baseline.
**HILLA:**

**Baseline:**
During all three sets in baseline there was barely any choice making. When limited choice making became apparent (set 2 sessions 1 and 4 or set 3 session 2), confirming the choice occurred seldom and choice making did not appear intentional.

**Intervention:**
Once the intervention began, set 1, session 1, there was a clear change. Hilla chose almost 100% of the time. She confirmed her choice a little less during set 1 (25/29 songs). During intervention set 2, session 1, the confirming of choice dropped drastically, but thereafter she almost always confirmed her choice, and this trend was apparent throughout set 2 when she confirmed 18/24 songs (75%). Her confirmation of choice of the songs increased in set 3 with 17/18 songs (94%).

Hilla scored high when choosing the first time as well as when confirming her choice. She was consistent throughout the intervention, which suggests that Hilla immediately understood the difference between baseline and intervention.

It was interesting to see that during session 1 in sets 2 and 3, Hilla had fallen somewhat behind when confirming her choice, but immediately thereafter she chose and confirmed her choice 100% of the time. The reason for falling behind during the first sessions may be due to the change in sets, which meant change in songs. Hilla’s favorite song (the Monkey) was in set 1. It could be that she was dissatisfied when the song did not appear during set 2 or 3.

**Baseline probes:**
The probes in sets 2 and 3 revealed that most of the time no confirmation of choice was made, thus choices cannot be assumed to be intentional during baseline probes.
In the first baseline probe of set 2, Hilla demonstrated high levels of choice making and confirmation of choice. In the second baseline probe of set 3 her choice making and confirmation of choice had dropped. It could be inferred that initially Hilla did not realize that she would not be hearing the song she had chosen and therefore continued to choose and to confirm her choice during the first probe.

**Maintenance probes:**

The intentional choice making ability seen during intervention sessions continued consistently during maintenance probes. In set 1, Hilla confirmed her choice 17/18 songs (94%), and in set 2 she confirmed her choice 5/6 songs (83%).

**Maintenance:**

During maintenance sessions Hilla confirmed her choice for 17/18 songs (94%), 18/18 songs (100%) and 18/18 (100%) in sets 1, 2, and 3 respectively.
Figure 4. Hilla –selection and confirmation of songs.

Baseline | Intervention | Maintenance

Set 1 # of songs selected in one session

Set 2 # of confirmed songs in one session

Set 3
Effect size calculation of Intervention and Maintenance compared with Baseline

**Baseline to intervention:**

Baseline Pre-test scores = 1 0 0 0 0 0 1 0 3 0 1  
Intervention Post-test scores = 5 4 5 5 6 2 5 5 6 5 6 6  
Mean pre-test scores = 0.50. Mean post-test scores = 5.00  
Standard deviation of pre-test scores = 0.90  
Effect size = (5.00 - 0.50)/ 0.90 = 5.00

**Baseline to maintenance:**

Baseline Pre-test scores = 1 0 0 0 0 0 1 0 3 0 1  
Maintenance Post-test scores = 5 6 6 6 6 6 6 6 6 6  
Mean pre-test scores = 0.50. Mean post-test scores = 5.89  
Standard deviation of pre-test scores = 0.90  
Effect size = (5.89 - 0.50)/ 0.90 = 5.99

The scores from both the intervention phase, and the maintenance phase when compared with the baseline scores show a very large effect, with an increased effect noted in the maintenance scores over baseline.
**MEIRAV:**

**Baseline:**
During baseline there was barely any choice making in set 1, choosing only 7/24 songs (29%). During baseline sets 2 and 3, the choice making was higher than in set 1 and resulted in 10/24 songs (42%) and 16/24 songs (67%) respectively. This low percentage of choice making followed by an even lower rate of intentional repetition (1/7 songs (14%), 6/10 songs (60%) and 8/16 songs (50%) – sets 1, 2 and 3 respectively).

**Intervention:**
During intervention there was a marked change. Every session, Meirav chose and confirmed her choice more than the previous session. In set 1, Meirav demonstrated intentional choice making in 24/26 songs (92%). During the first sessions in sets 2 and 3, the number of songs confirmed dropped, but from that point on repeated/intentional song choosing increased. In set 2, Meirav demonstrated intentional choice making in 17/24 opportunities (71%) and in set 3, 22/24 songs (92%). The number of times she confirmed her choice became almost equal to the number of initial choices during session two and throughout the sessions in sets 2 and 3.

The results of Meirav’s increasing consistency in choice making suggests that she was learning and adapting to the new method of choice, a change that seemed to occur when music was introduced.

**Baseline probes:**
The baseline probes in sets 2 and 3 were (40% & 23% respectively). Meirav showed no consistency in confirming her choice, thus choices cannot be assumed to be intentional during baseline probes.
**Maintenance probes:**

Meirav confirmed her choice 14/18 songs (78%) during maintenance probes in set 1 and 18/18 songs (100%) during set 2.

**Maintenance:**

During maintenance sessions Meirav revealed a ceiling effect (100%) in intentional choice making in all three sets.

**Figure 5. Meirav – Song selection and confirmation.**

![Graph showing song selection and confirmation over sessions](image-url)
**Effect size calculation of Intervention and Maintenance compared with Baseline**

**Baseline to intervention:**

Baseline Pre-test scores = 0 0 0 1 3 3 0 0 3 3 1 1  
Intervention Post-test scores = 3 4 5 6 6 2 5 5 4 6 6  
Mean pre-test scores = 1.25. Mean post-test scores = 4.75  
Standard deviation of pre-test scores = 1.36  
Effect size = (4.75 – 1.25)/ 1.36 = 2.57

**Baseline to maintenance:**

Baseline Pre-test scores = 0 0 0 1 3 3 0 0 3 3 1 1  
Maintenance Post-test scores = 6 6 6 6 6 6 6 6 6  
Mean pre-test scores = 1.25, Mean post-test scores = 6.00  
Standard deviation of pre-test scores = 1.36  
Effect size = (6.00 – 1.25)/ 1.36 = 3.49

The scores from both the intervention phase, and the maintenance phase when compared with the baseline scores show a very large effect, with an increased effect noted in the maintenance scores over baseline.
**RACHEL:**

**Baseline:**

The baseline in all three sets during choice making was very inconsistent: 60% in set 1, 38% in set 2, and 66% in set 3. The intentional choice making was very limited and stood at 55% in set 1, 18% in set 2 and 65% in set 3.

**Intervention:**

As soon as the intervention began (set 1, session 1) primary choice making and confirmation increased and stabilized. Choice making in Set 1 was high with 22/23 songs (96%). During set 2 the confirmation of choice was quite high (79%) although not as high or as stable as in set 1 and set 3 (87.5%).

Rachel showed a big difference between baseline and intervention. Since she was less experienced in choosing and probably had less difficulty changing from one method of choosing to the other, the new procedure was accepted immediately. As a participant, Rachel was the most ‘naïve’ of all seven participants, and therefore her responses can be assumed to be the purest example in this research. As she is an inexperienced chooser, her reaction can be attributed specifically to the intervention condition (songs).

**Baseline probes:**

The probes in sets 2 and 3 indicate a degree of influence from the intervention, as some confirmation of choices emerged in the baseline probes in set 2, and to a lesser degree in set 3, where confirmation of choice started to drop significantly.
**Maintenance probes:**

During set 1 intentional choice making resulted in 21/24 songs (87.5%) and during set 2 it resulted in 12/12 songs (100%).

**Maintenance:**

During all maintenance sessions, a high rate of intentional choice making was evident: 96% in sets 1 and 2, and 100% in set 3.

**Figure 6. Rachel –selection and confirmation of songs.**

![Graph showing selection and confirmation of songs in baseline, intervention, and maintenance phases across sets 1, 2, and 3.](image)
Effect size calculation of Intervention and Maintenance compared with Baseline:

**Baseline to intervention:**
Baseline Pre-test scores = 2 4 0 3 1 2 2 0 0 0 2 5 1 3 2
Intervention Post-test scores = 6 5 5 6 4 4 5 4 5 4 5 6 6
Mean pre-test scores = 1.80. Mean post-test scores = 5.00
Standard deviation of pre-test scores = 1.52
Effect size = (5.00 – 1.80) / 1.52 = 2.11

**Baseline to maintenance:**
Baseline Pre-test scores = 2 4 0 3 1 2 2 0 0 0 2 5 1 3 2
Maintenance Post-test scores = 5 6 6 6 6 6 5 6 6 6
Mean pre-test scores = 1.80, Mean post-test scores = 5.82
Standard deviation of pre-test scores = 1.52
Effect size = (5.82 – 1.80)/ 1.52 = 2.64

The scores from both the intervention phase, and the maintenance phase when compared with the baseline scores show a very large effect, with an increased effect noted in the maintenance scores over baseline.
**TALI:**

**Baseline:**
The baseline in sets 1 and 2 revealed a very low incidence of intentional choice making, however with a significant increase of confirmed choices in set 3. (22%, 20% and 74% in sets 1, 2 and 3 respectively).

**Intervention:**
During intervention set 1, session 1, there was very little choice making. From session 2 and thereafter, Tali chose 20/24 songs (83%), 29/30 songs (96%) and 18/18 songs (100%) in sets 1, 2 and 3 respectively. She confirmed the choice most of the time: 16/20 songs (80%), 25/29 songs (86) and 18/18 songs (100%) during sets 1, 2, and 3 respectively.

**Baseline probes:**
The baseline probes in sets 2 and 3 revealed higher percentages of confirming her choice than expected (55% and 48%), when compared with percentages found in other participants during baseline probe.

**Maintenance probes:**
During set 1 Tali confirmed her choice 16/18 songs (89%) and 6/6 songs (100%) of the time during set 2.

**Maintenance:**
In the maintenance sessions Tali chose and confirmed her choice 95%-100% of the time.
Figure 7. Tali – song selection and confirmation.

Baseline | Intervention | Maintenance

# of songs selected in one session
# of confirmed songs in one session
Effect size calculation of Intervention and Maintenance compared with Baseline

Baseline to intervention:
Baseline Pre-test scores = 0 0 1 1 0 0 1 1 0 1 4 3 2 2 3
Intervention Post-test scores = 0 6 5 5 4 4 5 6 6 6 6 6
Mean pre-test scores = 1.27. Mean post-test scores = 4.92
Standard deviation of pre-test scores = 1.28
Effect size = (4.92 – 1.27)/ 1.28 = 2.85

Baseline to maintenance:
Baseline Pre-test scores = 0 0 1 1 0 0 1 1 0 1 4 3 2 2 3
Maintenance Post-test scores = 6 6 6 5 6 6 6 6 6
Mean pre-test scores = 1.27. Mean post-test scores = 5.89
Standard deviation of pre-test scores = 1.28
Effect size = (5.89 – 1.27)/ 1.28 = 3.61

The scores from both the intervention phase, and the maintenance phase when compared with the baseline scores show a very large effect, with an increased effect noted in the maintenance scores over baseline.

TALIA:
Baseline:
During baseline Talia showed low percentage of choosing, which resulted in: 6/24 (25%), 12/24 (50%) and 13/24 (54%) in sets 1, 2 and 3 respectively. However her confirmation of her choice in sets 1 and 3 was at a much higher ratio when compared with other participants: 4/6 (66%); and 11/13 (85%) in sets 1, 2 and 3 respectively. In set 2 there was a marked decrease in confirmation of choice with only 2/12 (17%),
**Intervention:**

During intervention set 1, Talia’s choice making improved and resulted in 38/42 songs (91%), 16/18 songs (89%), and 16/18 songs (89%) in sets 1, 2, and 3 respectively. Although sessions 1-3 in set 1 showed slight inconsistencies, by the fourth session, set 1 and onwards, she was able to confirm her choice with a higher degree of consistency.

**Baseline probes:**

The baseline probes in set 2 and 3 revealed a medium to low confirmation of choice making standing at 4/10 songs (40%); and 6/17 songs (35%) in sets 1 and 2 respectively. Therefore, some intentional choice making was evident during the probes.

**Maintenance probes:**

Talia confirmed her choices in set 1, 11/12 songs (92%) and 5/6 songs (83%) during set 2. A very consistent and high intentional choice making was demonstrated during maintenance probes.

**Maintenance:**

Talia sustained a high percentage of intentional choice making during maintenance sessions. This included 18/18 (100%), 16/18 (89%) and 17/18 (94%) in sets 1, 2, and 3 respectively.
Figure 8. Talia –selection and confirmation of songs.

Baseline      Intervention                 Maintenance

0 1 2 3 4 5 6
Sessions

Set 1

Set 2

Set 3

0 1 2 3 4 5 6 7 8 9 10 11 12 13 14 15 16 17 18 19 20 21 22

# of songs selected in one session

# of songs confirmed in one session
Effect size calculation of Intervention and Maintenance compared with Baseline

**Baseline to intervention:**

Baseline Pre-test scores = 0 3 1 0 1 1 0 0 4 5 1 1  
Intervention Post-test scores = 4 3 3 4 5 5 5 5 6 5 6 5  
Mean pre-test scores = 1.42, Mean post-test scores = 4.69  
Standard deviation of pre-test scores = 1.68  
Effect size = \( \frac{4.69 - 1.42}{1.68} = 1.95 \)

**Baseline to maintenance:**

Baseline Pre-test scores = 0 3 1 0 1 1 0 0 4 5 1 1  
Maintenance Post-test scores = 6 6 6 5 5 6 5 6 6  
Mean pre-test scores = 1.42, Mean post-test scores = 5.67  
Standard deviation of pre-test scores = 1.68  
Effect size = \( \frac{5.67 - 1.42}{1.68} = 2.53 \)

The scores from both the intervention phase, and the maintenance phase when compared with the baseline scores show a very large effect, with an increased effect noted in the maintenance scores over baseline.
3.1.2 Intentional choice making - group

During baseline all girls chose inconsistently and while the mean results indicate generally low intentional choice there were also differences between girls and between sets. Variability in intentional choice making can be observed on Table 5.

Table 5. Intentional choice making during baseline in percentages.

<table>
<thead>
<tr>
<th>Child</th>
<th>Set #</th>
<th>Aviv</th>
<th>Elisheva</th>
<th>Hilla</th>
<th>Meirav</th>
<th>Rachel</th>
<th>Tali</th>
<th>Talia</th>
<th>Average</th>
</tr>
</thead>
<tbody>
<tr>
<td>Set 1</td>
<td>18</td>
<td>38</td>
<td>20</td>
<td>14</td>
<td>55</td>
<td>22</td>
<td>66</td>
<td></td>
<td>33</td>
</tr>
<tr>
<td>Set 2</td>
<td>21</td>
<td>38</td>
<td>9</td>
<td>60</td>
<td>18</td>
<td>20</td>
<td>17</td>
<td></td>
<td>26</td>
</tr>
<tr>
<td>Set 3</td>
<td>15</td>
<td>27</td>
<td>36</td>
<td>50</td>
<td>65</td>
<td>74</td>
<td>85</td>
<td></td>
<td>50</td>
</tr>
</tbody>
</table>

No apparent trends for improvement or deterioration can be seen during intentional choice making at baseline. In some participants the incidence was rising and in others was dropping. During baseline probe sessions this lack of change was seen again as all girls more typically presented low rates of intentional choice making. Some girls showed an increase in intentional choice making and others a decrease. This inconsistency can be viewed in Table 6.

Table 6. Intentional choice making during probes in percentages.

<table>
<thead>
<tr>
<th>Child</th>
<th>Set #</th>
<th>Aviv</th>
<th>Elisheva</th>
<th>Hilla</th>
<th>Meirav</th>
<th>Rachel</th>
<th>Tali</th>
<th>Talia</th>
<th>Average</th>
</tr>
</thead>
<tbody>
<tr>
<td>Set 2</td>
<td>30</td>
<td>22</td>
<td>66</td>
<td>40</td>
<td>32</td>
<td>55</td>
<td>40</td>
<td></td>
<td>41</td>
</tr>
<tr>
<td>Set 3</td>
<td>14</td>
<td>60</td>
<td>44</td>
<td>23</td>
<td>32</td>
<td>48</td>
<td>35</td>
<td></td>
<td>37</td>
</tr>
</tbody>
</table>

Both baseline and probe sessions rated below 50% during intentional choice making. Once the intervention began, all girls began choosing much more consistently and the rates of intentional choice making, demonstrated by confirmation of choices, also
rose. Most girls confirmed their choice within the first session of set 1, while others within the first 3 sessions. Looking at Table 7, it is clear that intentional choice making was apparent to a high degree in all girls with Rett syndrome who participated in the research. Average intentional choice making rose to levels over 83%.

Table 7. Intentional choice making during intervention in percentages.

<table>
<thead>
<tr>
<th>Child</th>
<th>Set #</th>
<th>Aviv</th>
<th>Elisheva</th>
<th>Hilla</th>
<th>Meirav</th>
<th>Rachel</th>
<th>Tali</th>
<th>Talia</th>
<th>Average</th>
</tr>
</thead>
<tbody>
<tr>
<td>Set 1</td>
<td></td>
<td>66</td>
<td>83</td>
<td>86</td>
<td>92</td>
<td>96</td>
<td>80</td>
<td>91</td>
<td>85</td>
</tr>
<tr>
<td>Set 2</td>
<td></td>
<td>83</td>
<td>94</td>
<td>75</td>
<td>71</td>
<td>79</td>
<td>86</td>
<td>89</td>
<td>83</td>
</tr>
<tr>
<td>Set 3</td>
<td></td>
<td>100</td>
<td>96</td>
<td>94</td>
<td>92</td>
<td>88</td>
<td>100</td>
<td>89</td>
<td>94</td>
</tr>
</tbody>
</table>

During the maintenance sessions all girls made clear intentional choice making and confirmed their first choices continuously.

Table 8. Intentional choice making during maintenance in percentages.

<table>
<thead>
<tr>
<th>Child</th>
<th>Set #</th>
<th>Aviv</th>
<th>Elisheva</th>
<th>Hilla</th>
<th>Meirav</th>
<th>Rachel</th>
<th>Tali</th>
<th>Talia</th>
<th>Average</th>
</tr>
</thead>
<tbody>
<tr>
<td>Set 1</td>
<td></td>
<td>100</td>
<td>92</td>
<td>94</td>
<td>100</td>
<td>96</td>
<td>95</td>
<td>100</td>
<td>97</td>
</tr>
<tr>
<td>Set 2</td>
<td></td>
<td>100</td>
<td>100</td>
<td>100</td>
<td>100</td>
<td>96</td>
<td>100</td>
<td>89</td>
<td>98</td>
</tr>
<tr>
<td>Set 3</td>
<td></td>
<td>100</td>
<td>100</td>
<td>100</td>
<td>100</td>
<td>100</td>
<td>100</td>
<td>94</td>
<td>99</td>
</tr>
</tbody>
</table>

Table 8 clearly represents a slow but constant rise in the intentional choice making rate from 94% at intervention sessions to 97% at the beginning of the maintenance part gradually growing to the impressive 99% of intentional choice making. Viewing at the data from the first time the songs were presented to all the participants as a
group, and up to the last maintenance session, one can see that change has occurred as well as when this change took place.

Average intentional choice making during baseline was 36%; average intentional choice making during probe was 39%. During intervention average intentional choice making was 87.5%. During maintenance probe sessions 90% and during maintenance sessions, average intentional choice making was 98%. See Figure 8.

**Group Effects within sets**

**Set 1**

**Baseline to intervention:**

Mean Pre-test baseline scores = 1 0.6 0.25 0.25 2 0.4 1

Mean Post-test Intervention scores = 3.83 3.8 5 4.8 5.5 4 4.14

Mean pre-test = 0.79, Mean post-test = 4.44

Standard deviation of pre-test mean scores = 0.62

Effect size = \( \frac{4.44 - 0.79}{0.62} = 5.89 \)

**Baseline to maintenance:**

Mean Pre-test baseline scores = 1 0.6 0.25 0.25 2 0.4 1

Mean Post-test Maintenance scores = 5 5.5 5.67 6 5.75 6 6

Mean pre-test = 0.79, Mean post-test = 5.7

Standard deviation of pre-test mean scores = 0.62

Effect size = \( \frac{5.7 - 0.79}{0.62} = 7.92 \)

**Set 2**

**Baseline to intervention:**

Mean Pre-test Baseline Scores = 1.2 1 0.25 1.5 0.8 0.6 0.5

Mean Post-test Intervention Scores = 5 5.33 4.5 4.25 4.5 5 5.33

Mean pre-test = 0.84, Mean post-test = 4.84

Standard deviation of pre-test mean scores = 0.43

Effect size = \( \frac{4.84 - 0.84}{0.43} = 9.3 \)
**Baseline to maintenance:**
Mean Pre-test baseline scores = 1.2 1 0.25 1.5 0.8 0.6 0.5
Mean Post-test Maintenance scores = 6 6 6 5.75 5.67 5.33
Mean pre-test = 0.84. Mean post-test = 5.82
Standard deviation of pre-test mean scores = 0.43
Effect size = (5.82 - 0.84)/ 0.43 = **11.58**

**Set 3**

**Baseline to intervention:**
Mean Pre-test baseline scores = 0.6 0.6 1 2 2.6 2.8 2.75
Mean Post-test Intervention scores =
6 5.25 5.67 5.33 5.25 6 5.33
Mean pre-test = 1.76. Mean post-test = 5.55
Standard deviation of pre-test mean scores = 1.01
Effect size = (5.55 - 1.76)/ 1.01 = **3.75**

**Baseline to maintenance:**
Mean Pre-test Baseline scores = 0.6 0.6 1 2 2.6 2.8 2.75
Mean Post-test maintenance scores = 6 5.67 6 6 6 5.67
Mean pre-test = 1.76. Mean post-test = 5.91
Standard deviation of pre-test mean scores = 1.01
Effect size = (5.91 - 1.76)/ 1.01 = **4.11**

Calculating the group mean scores for sets 1, 2, and 3 reveal differences in effects between sets. Very large effect sizes were scored for differences between baseline and intervention, and baseline and maintenance in all three sets. However, while larger differences in effect is noted when comparing the baseline with both intervention and maintenance in Set 1 (5.89 & 7.92) and Set 2 (9.3 & 11.98), in set 3 there is a reduction of effect (3.75 & 4.11), which may be due to increased mean
scores in subjects five, six and seven during baseline. The effect size calculation also reflects a larger effect in the difference between baseline and maintenance than between baseline and intervention in all three sets of songs. This indicates that the girls not only demonstrated retained, but actually improved choosing responses after a period of no intervention (two week, four week and six week gaps).

**Total Effect size for the whole group**

**Baseline to intervention:**
Mean Pre-test scores = 0.93 0.73 0.5 1.25 1.8 1.27 1.42
Mean Post-test scores = 4.69 4.67 5 4.75 5 4.92 4.69
Mean pre-test = 1.13. Mean post-test = 4.82
Standard deviation of pre-test scores = 0.44
Effect size = (4.82 - 1.13)/ 0.44 = 8.39

**Baseline to maintenance:**
Mean Pre-test scores = 0.93 0.73 0.5 1.25 1.8 1.27 1.42
Mean Post-test scores = 5.67 5.73 5.89 6 5.82 5.89 5.67
Mean pre-test = 1.13. Mean post-test = 5.81
Standard deviation of pre-test scores = 0.44
Effect size = (5.81 - 1.13)/ 0.44 = 10.64

The overall effect size on the mean scores of the whole group of seven participants, calculated on the mean scores from all three sets together, demonstrate very large effect sizes, and again emphasis a larger overall effect during the maintenance phase than was found in the intervention phase when compared with the baseline.
Figure 9. Average percentage of intentional choice making during different parts of the research.
3.2 RESPONSE TIME

Introduction

Picture symbols or orthographies of songs were presented in the study as stimulus. A five steps choice making procedure was implemented as the stimulus was presented and the duration of each step was 15 seconds. Steps one and two were considered as independent choice making, while steps three to five all involved some degree of cues, prompting and assistance - step three, least prompting and step five, most prompting (more detailed explanation in the procedure section in the method chapter).

The following Figures 10-17 (for each participant), will illustrate four sessions out of the intervention period all from set one. The intervention period was selected in order to analyze response time, because by that time the participants had become familiar with the choosing procedure. Their motivation for choosing was at its highest during intervention as they were receiving the song for their response.

Each session on the graph includes five data points, indicating the song stimuli presented. The participant had the opportunity to respond up to five times (or steps). The steps were measured by 15 seconds each, represented on the y-axis of the graph. If they responded by step one, they responded within 15 seconds to the stimulus. If it took longer than 15 seconds and up to 30 seconds it was considered as step two. This was indicated where data points were entered against the step marked 30 seconds.

The response time was carefully monitored for the first three to four sessions. After that, the girls had begun to consistently respond within 15 seconds.

Some of the following Figures show three sessions, and some show four sessions. This was due to the fact for some participants it took longer for the results to become consistent within the 15 seconds. When no response was observed within the total monitored time for response of 75 seconds, it was indicated in the Figures with an open gap between the stimuli.
All of the following Figures indicate response time within the five steps (15 seconds each step). Because all participants’ response time was within the range of 15 seconds by the third or fourth session, only one Figure was chosen to demonstrate the exact time analysis.

3.2.1 Response time in individual cases

**AVIV**

During the first and second session, Aviv’s response times when presented with the stimulus ranged from \( \leq 15 \) to \( \leq 30 \) seconds averaging 13 sec. and 27.5 sec. In sessions one and two respectively. Starting from the beginning of the third session she was showing consistent response within the 15 seconds, averaging 15 sec. or less at third and fourth sessions. See Figure 10.

**Figure 10.** Aviv’s response time to stimulus.
A sample graph showing the Actual Response Time is presented for the first participant only - **Figure 11**.

**Figure 11. Actual response time to stimulus.**

The blue line represents the lowest time frame possible (first choice making step according to the research protocol). The graph shows that it took Aviv two sessions to reach consecutive short response time. Out of the 24 presentations of stimuli, 14 are under 15 seconds. Average time for responding to a stimulus during all 4 sessions was 10.7 seconds, while average response time to the stimulus at sessions 3-4 and onward was stable at around 5.3 seconds.

Thereafter, the data for the other participants will be presented showing their response time to stimulus over four (or five) sessions. These graphs relate to “choice - making stages” (15 seconds each), and are documented here to illustrate changes in response time primarily.
Figure 11 indicates the same information as previously recorded in Aviv’s graph and is set out in the same way as described in the introduction section.

During the first session, set 1, the response time to the stimulus ranged between $\leq 15$ to $\leq 45$ seconds averaging at 37.5 sec. During the second session, the response time was $\leq 15$ to $\leq 45$ seconds averaging at 42.5, and twice there was no response at all (calculated at a value of 75 seconds). Towards the end of the second session and until the middle of the third session, the response time remained within a range of $\leq 15$ to $\leq 45$ seconds, but the average response time dropped to 30 seconds. In the middle of the third session, and thereafter, the response time ranged between $\leq 15$ to $\leq 30$ seconds, with a mean of 20 seconds.

Figure 12. Elisheva’s response time to stimulus.

Elisheva’s response time to the first and second stimulus during the second session fell behind and she did not respond even after the fifth step. This may have been due to the circumstance that she had been crying in her classroom prior to the session. Her state of being upset could have set her back and interfered with her response. By the third choice of the same session, she responded very rapidly.
**HILLA**

Figure 13 shows that during the first session, reaction time to the stimulus ranged between $\leq 15 - \leq 45$ seconds averaging at 22.5 sec. During the second session, the response time was reduced, ranged between $\leq 15 - \leq 30$ seconds, and averaged at 17.5 sec. During the middle of the second session and thereafter (sessions 2 and 3) the response time was 15 seconds and less, each time the stimulus was presented.

**Figure 13. Hilla’s response time to stimulus.**

Hilla’s response time was 15 seconds or less within the second session. She was eager to choose during set 1, as she was very familiar with four out of the six songs, which she happily chose. The Monkey song was her very favorite, and her response time when choosing the Monkey song was faster than when choosing any other songs.

**MEIRAV**

During the first session, response time to the stimulus ranged between 15 to $\leq 45$ seconds and averaged at 30 seconds. Towards the middle of the second session and thereafter, the response time ranged between 15 to $\leq 30$ seconds. The average
response time to stimulus in the second session was 22.5 seconds and 20 seconds in the third session. In the fourth session and onwards, the reaction time was always 15 seconds or less. See Figure 14.

**Figure 14. Meirav’s response time to stimulus.**

![Response Time Graph](image)

**RACHEL**

Figure 14 reveals that during the first session, response time to the stimulus ranged between 15 - ≤ 30 seconds. During the second session, the response time ranged between 15 - ≤ 45 seconds. During the third session and thereafter, the response time was ≤ 15 seconds.

Rachel’s response time was 15 seconds or less during the first session. Her response time increased during the second session and ranged between 15-45 seconds. The slower response time may have been due to Rachel’s illness a few days prior to that session. She returned to her maximum performance from session three, and thereafter. See Figure 15.
TALI

During the first session, response time to the stimulus ranged between ≤ 15 to ≤ 60 seconds, and averaged at 37.5 seconds. In the second session, the response time dropped, ranging between 15 to ≤ 30 seconds, averaging at 25 seconds. From the third session and thereafter, the response time was ≤ 15 seconds. See Figure 16.

Figure 15. Rachel’s response time to stimulus.

Figure 16. Tali’s response time to stimulus.
TALIA

During the first session the response time to the stimulus ranged between $\leq 15$ to $\leq 75$ seconds. One time Talia did not respond at all. During session two and three, response time was between $\leq 15$ - $\leq 30$ seconds. During the fourth session and thereafter, the response time was $\leq 15$ seconds.

Talia’s response time was very inconsistent during the first session. She was extremely excited and seemed not sure of what was expected of her. It became quite consistent during the second session with response time of 15-30 seconds.

Figure 17. Talia’s response time to stimulus.
3.2.2 Response time as a group

The response time during each trial throughout the research was measured at 15 seconds intervals. In all graphs, the participants typically reveal delayed responses during the first two sessions. Once they had learned the procedure of making a choice when presented with the song stimulus, their choice making became very consistent and response time dropped considerably. Most participants needed at least 15 seconds and on some occasion more than 75 seconds when making a choice during the first 2-3 sessions. Thereafter, the response time dropped dramatically in all cases. At that time they needed ≤ 15 - ≤ 30 seconds to respond to the stimulus.

**Figure 11** is a sample of time analysis of true response time for one participant. All other girls match this sample graph and the process shown in it, and therefore it was unnecessary to continue with this level of detailed analysis of true response time for all the participants.

**Figure 18** reveals the average response time for all participants during the first four sessions.

**Figure 18. Average response time - all participants.**
The changes in response time over sessions 1-4 can be presented in a different graph including Standard Deviation as indicated in Figure 19.

Figure 19. Development of average response time over sessions.

![Figure 19](image)

Figure 19 demonstrates that together with the decline in response time, Standard Deviation or the differences between the participants was reduced, revealing increasing homogeneity in the scores of response time.

In sessions 1 and 2 the Standard Deviation was at 17 and 14 seconds respectively, whereas in session 3 it dropped to a value of 8. In session 4, the standard deviation stood at a value of 0.9.
3.3 LEARNING PROCESS  
(Within sets, between sets and during maintenance sessions)

Introduction

Learning process occurred in all the participants and can be viewed on three levels. The first level of learning occurred within the first set. There was an ascending trend, showing that learning took place within the first few sessions during the first set. The participants learned a new procedure of intentional choice making, reaching the criterion that was established for that learning to be considered consistently reliable.

The second level of learning took place in the number of sessions needed for the participants to fulfill the criteria of moving from one set of symbols to the next. The number of required sessions typically reduced in most participants as the intervention progressed.

The third level of learning can be seen during maintenance sessions. The participants’ long term memory was evident as they remembered the procedure of choice making, and their preferred songs.
3.3.1 Learning process in individual cases

AVIV

Within sets: Table 9 illustrates the process of learning demonstrated by the increasing consistency of Aviv’s confirmation of choices.

Table 9. Aviv’s trend of learning within sets.

<table>
<thead>
<tr>
<th>Sets</th>
<th>Set 1</th>
<th>Set 2</th>
<th>Set 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Session 1</td>
<td>3/6</td>
<td>4/6</td>
<td>6/6</td>
</tr>
<tr>
<td>Session 2</td>
<td>2/6</td>
<td>5/6</td>
<td>6/6</td>
</tr>
<tr>
<td>Session 3</td>
<td>3/6</td>
<td>6/6</td>
<td>6/6</td>
</tr>
<tr>
<td>Session 4</td>
<td>5/6</td>
<td>5/6</td>
<td>XXXX</td>
</tr>
<tr>
<td>Session 5</td>
<td>5/6</td>
<td>XXXX</td>
<td>XXXX</td>
</tr>
<tr>
<td>Session 6</td>
<td>5/6</td>
<td>XXXX</td>
<td>XXXX</td>
</tr>
</tbody>
</table>

Aviv's ability to make choices during the first session was clear. Confirming her choice was limited and became compatible with her first choice only during the fourth session set 1. Aviv was evidently learning effective choice making over time.

Between sets: It took Aviv six sessions in set 1 to fulfill the criteria, four sessions to complete set 2, and three sessions to complete set 3. Learning occurred between each set as well as the number of sessions between one set to the next was reduced.

Maintenance sessions: During first maintenance session set 1, Aviv confirmed her choice 5 out of 6 times. In all the remaining sets, she confirmed her choice 6 out of 6 times.
ELISHEVA

Within sets (see table 10’): An ascending trend of learning took place and was demonstrated in set 1. Elisheva revealed an increasing consistency of her confirmation of choices.

Table 10. Elisheva’s trend of learning within sets.

<table>
<thead>
<tr>
<th>Sets</th>
<th>Set 1</th>
<th>Set 2</th>
<th>Set 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Session 1</td>
<td>2/5</td>
<td>5/6</td>
<td>5/6</td>
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</tr>
<tr>
<td>Session 3</td>
<td>5/5</td>
<td>6/6</td>
<td>6/6</td>
</tr>
<tr>
<td>Session 4</td>
<td>5/6</td>
<td>XXXX</td>
<td>6/6</td>
</tr>
<tr>
<td>Session 5</td>
<td>5/6</td>
<td>XXXX</td>
<td>XXXX</td>
</tr>
</tbody>
</table>
**Between sets:** It took Elisheva six sessions to fulfill the criteria in set 1, three sessions to complete set 2, and four sessions to complete set 3. Learning occurred between set 1 and set 2 and set 1 and set 3. The duration of moving from set 1 to the next (set 2) was reduced.

**Maintenance sessions:** During session one, sets 1 and 3, Elisheva confirmed her choice 5 out of 6 times as well as in session 3, set 1. In all other sets she confirmed her choice 6 out of 6 times.

---

**Figure 21: Elisheva - learning process**

![Graph showing learning process](image)

**Index:**
- # of songs selected in one session
- # of confirmed songs in one session
- # of sessions to complete a criteria
**HILLA**

**Within sets:** Hilla also demonstrated an increasing ability to confirm her choices during set 1, sustained in sets 2 and 3.

**Table 11. Hilla’s trend of learning within sets.**

<table>
<thead>
<tr>
<th>Sets</th>
<th>Set 1</th>
<th>Set 2</th>
<th>Set 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Session 1</td>
<td>5/5</td>
<td>2/6</td>
<td>5/6</td>
</tr>
<tr>
<td>Session 2</td>
<td>4/6</td>
<td>5/6</td>
<td>6/6</td>
</tr>
<tr>
<td>Session 3</td>
<td>5/6</td>
<td>5/6</td>
<td>6/6</td>
</tr>
<tr>
<td>Session 4</td>
<td>6/6</td>
<td>6/6</td>
<td>XXXX</td>
</tr>
<tr>
<td>Session 5</td>
<td>6/6</td>
<td>XXXX</td>
<td>XXXX</td>
</tr>
</tbody>
</table>

**Between sets:** It took Hilla five sessions to fulfill the criterion in set 1, four sessions to fulfill set 2 and three sessions in set 3. Learning occurred between each set. The duration of moving from one set to the next was reduced.

**Maintenance:** In the first session set 1 of maintenance, Hilla confirmed her choice 5 out of 6 times. During all other maintenance sessions (in all sets), she confirmed her choice 6 out of 6 times. Memory was kept and learning was maintained.
Within sets: As found with the other participants, there is an increase in confirmation of choice in set 1, which is well sustained in sets 2 and 3 (Table 12).

In the first session set 2, Meirav confirmed her choice 2 out of 6 times, then confirming her choice 5 out of 6 times during sessions: two, three and four.

In the first session set 3, Meirav confirmed her choice 4 out 6 times, then confirming her choice 6 out of 6 times during sessions: two, three and four. Learning had occurred.
Table 12. Meirav’s trend of learning within sets.

<table>
<thead>
<tr>
<th>Sets</th>
<th>Set 1</th>
<th>Set 2</th>
<th>Set 3</th>
</tr>
</thead>
<tbody>
<tr>
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<td>4/5</td>
<td>5/6</td>
<td>6/6</td>
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<tr>
<td>Session 3</td>
<td>5/5</td>
<td>5/6</td>
<td>6/6</td>
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<tr>
<td>Session 4</td>
<td>6/6</td>
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</tr>
<tr>
<td>Session 5</td>
<td>6/6</td>
<td>XXXX</td>
<td>XXXX</td>
</tr>
</tbody>
</table>

**Between sets:** It took Meirav five sessions to fulfill the criteria in set 1, four sessions in set 2 and four sessions in set 3. Learning occurred between the first and the remaining sets. The duration of moving from set 1 to set 2 was reduced while set 3 stayed the same as set 2.

**Maintenance:** During all maintenance sessions in all sets, Meirav chose and confirmed her choice 6 out of 6 times. Learning was maintained.
Rachel's trend of learning within sets.

Table 13 shows that Rachel confirmed her choice consistently, and was learning choice making as the sessions proceeded.

**Table 13. Rachel’s trend of learning within sets.**

<table>
<thead>
<tr>
<th>Sets</th>
<th>Set 1</th>
<th>Set 2</th>
<th>Set 3</th>
</tr>
</thead>
<tbody>
<tr>
<td>Session 1</td>
<td>6/6</td>
<td>4/5</td>
<td>4/6</td>
</tr>
<tr>
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<tr>
<td>Session 6</td>
<td>XXXX</td>
<td>6/6</td>
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</tr>
</tbody>
</table>
**Between sets:** Rachel reached the criterion of set 1 in four sessions. There was an increase in number of sessions in set two, completing the criterion in six sessions. During set 3 she completed the set in 4 sessions.

**Maintenance:** During all maintenance sessions in all sets, Rachel chose and confirmed her choice 6 out of 6 times (accept for session three, set 2 where she confirmed her choice 5 out of 6 times). Learning maintained during the maintenance sessions.

**Figure 24: Rachel - learning process**
**TALI**

**Within sets (see Table 14):** In the first session set 1, Tali confirmed 0 out of the 2 choices she had made. In session two, she confirmed 5 out 6 times and during sessions three and four she confirmed 5 out of 6 times. There was a big leap between, session one and session two in set 1. Learning had taken place between the two sessions.

Sets 2 and 3 showed an ascending trend over time, and clearly demonstrate Tali’s learning (see Table 14).

Table 14. Tali’s trend of learning within sets.

<table>
<thead>
<tr>
<th></th>
<th>Set 1</th>
<th>Set 2</th>
<th>Set 3</th>
</tr>
</thead>
<tbody>
<tr>
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<tr>
<td>Session 5</td>
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</tbody>
</table>

**Between sets:** Learning took place between sets, especially between sets 2 and 3. It took Tali four sessions to reach the criterion in set 1, five sessions in set 2 and three sessions in set 3.

**Maintenance:** Throughout maintenance sessions in all sets, Tali chose and confirmed her choice 6 out of 6 times (except for session one set 2, where she had confirmed her choice 5 out of 6 times). Learning maintained during the maintenance sessions.
**Figure 25: Tali - learning process**

**Table 15. Talia’s trend of learning within sets.**

<table>
<thead>
<tr>
<th>Sets</th>
<th>Set 1</th>
<th>Set 2</th>
<th>Set 3</th>
</tr>
</thead>
<tbody>
<tr>
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<td>Session 5</td>
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<tr>
<td>Session 7</td>
<td>5/6</td>
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</tr>
</tbody>
</table>
**Between sets:** Learning took place between sets. It took seven sessions to fulfill the criterion during set 1, three session in set 2, and three sessions in set 3. The number of sessions was drastically reduced between sets 1 and 2.

**Maintenance:** Throughout all three maintenance sessions in set 1, Talia confirmed her choice 6 out of 6 times. In set 2 she confirmed her choice 5 out of 6 times in sessions one and two, and 6 out of 6 times in session three. During set 3 she confirmed her choice 5 out of 6 times in session one and 6 out of 6 times in sessions two and three. Learning sustained during maintenance sessions.

**Figure 26: Talia - learning process**

![Graph showing learning process]

- **Index:**
  - # of songs selected in one session
  - # of confirmed songs in a session

- **# of sessions to complete a criteria**
3.3.2 Learning process as a group

Learning occurred within all participants. Most graphs show clearly that all participants had ascending trend when making intentional choices. During the first session in sets 2 and 3 the number of choices and confirmations of choice had dropped in most participants and climbed thereafter. The number of sessions to complete the pre-established criterion was reduced between the sets. This was true with most of the girls, thus learning took place between sets. All participants sustained a very high level of learning during maintenance sessions. The learning process of recognition and choice making of symbols had a long-term effect and was sustained almost three months after the termination of the intervention.
3.4 CHOICE PREFERENCE OF SONGS

Introduction

Eighteen songs were presented during this research: Twelve familiar and six unfamiliar songs. The familiar songs were the Nut, Monkey, Spider, Bee, Farm, Bird, Butterfly, Frog, Tree, Turtle, Fish and Elephant. The non-familiar songs were: The Train, Chick, Hands, Star, Crocodile and Rabbit. The number of times the songs were selected by the girls in comparison to the number of times the songs were presented, was calculated in percentage. The percentage of times the songs were selected reveal the song preference in each individual participant.

The participants chose one song out of two or four song selections (as indicated in the procedure section in the method chapter).

Figure 27 places the songs in order of preference, according to the mean scores from all participants. The most and least selected songs are referred to as most preferred and least-preferred songs.
There is a large gap between the five most and five least selected songs. The average percentage of choice of the five most selected songs was 70.6%, while the average percentage of choice of the five least preferred songs was 20.6%. The remaining eight songs can be grouped into two bands. The average percentage of the higher four songs (out of the eight) was 50% and the average percentage of the lower four songs was 35%.

3.4.1 Song preference in individual cases

Song preference in each individual participant was determined in percentage (Figures 27-34) and all songs were labeled in the same order. The five most and least selected songs will be referred to in each individual case.

AVIV

Most selected songs were: Train, Spider, Monkey, Bee and Nut, and the percentages of the number of times Aviv chose these songs are illustrated in Figure 28.

Aviv chose the Train song every time the song showed up (100%). Even though the song was a new one, she seemed to like it instantly. She laughed when she chose the song, got out of her seat, moved her body with large movement, spun around and returned to her seat.
The least selected songs were: Rabbit, Hands, Fish, Chick and Star, and the percentages of the number of times Aviv chose these songs is illustrated in Figure 28. The Fish song was the only familiar song out of these five songs.

Aviv’s least favorite song was the Rabbit song (0%). Although she never selected the song intentionally, she did select it at one time with prompting by the researcher. At that time she turned her head away, and walked out of her seat towards the door. Aviv was familiar with the Fish song and used to enjoy the song in the past when she was younger. She would typically smile and laugh when she heard it. This can indicate that she had outgrown the Fish song and has now other songs she would rather hear.

Figure 28. Aviv’s song selection.

ELISHEVA

The most selected songs were: Bee, Spider, Chick, Nut and Bird, and the percentages of the number of times Elisheva chose these songs is illustrated in
The Chick song was the only unfamiliar song out of the five most selected songs.

Elisheva’s favorite song was the Bee song (94%). She would act shyly when she chose this song by smiling looked at the floor, then looked up at the researcher and continued to smile.

Least selected songs were: Elephant, Crocodile, Hands, and Butterfly, and the percentages of the number of times Elisheva chose these songs are illustrated in. The Elephant and Butterfly songs were the only familiar song out of these five songs. Elisheva’s least preferred song was the Rabbit song (6%) as well. She chose it once and never returned to that song. Elisheva did not smile during the song and turned her head away.

Figure 29. Elisheva’s song selection.
The most selected songs were Monkey, Nut, Bee, Train and Spider, and the percentages of the number of times Hilla chose these songs are illustrated in (Figure 30). The Train song was the only unfamiliar song out of the five songs.

Hilla’s most favorite was the Monkey song (91%). The researcher can recall the Monkey song as Hilla’s favorite song long before this study had begun. Hilla would typically move her hands to the beat of the music. She stopped the hand movements exactly on the pauses of the music, leaning her hands on her abdomen and restart the hand movement once the next verse began, smiling throughout the song. Her response time was faster when she chose the monkey song, her activity level was higher and there was more vitality in her behavior.

The least selected songs were: Fish, Crocodile, Rabbit, Star and Hands, and the percentages of the number of times Hilla chose these songs is illustrated in (Figure 30). The Fish song was the only familiar song out of these five songs. Hilla’s least preferred song was the Fish song (8%). She was familiar with the Fish song and used to enjoy the song in the past when she was younger. This song is more like a cradle- song, and definitely more appropriate for younger children. She sat quietly, looked at the researcher with a serious face while the song was sung.
MEIRAV

The most selected songs were: Spider, Train, Monkey, Nut and Chick/Farm (same %), and the percentages of the number of times Meirav chose these songs is illustrated in Figure 31. The Train and Chick songs were unfamiliar songs.

Meirav’s favorite song, the Spider song (79%), does not have as high percentage (for a favorite). She had a variety of songs with a similar percentage score, and there was no one song that attracted her highest attention, as was clearly not the case for the six other participants. Meirav tried out different songs, but at the same time was very excited when she heard the Spider song.

The least selected songs were: Rabbit, Hands, Fish, Crocodile, and Butterfly and the percentages of the number of times Meirav chose these songs is illustrated in Figure 31. The Fish and Butterfly songs were the only familiar songs out of these five songs.
Meirav’s least preferred song was the Rabbit song (4%), which she had chosen only once. She began bellowing when the song was sung, a behavior quite similar to the one she exhibited during baseline (no song).

Figure 31. Meirav’s song selection.
RACHEL

The most selected songs were: Nut, Farm, Train, Bee/Bird (same %) and Monkey, and the percentages of the number of times Rachel chose these songs is illustrated in Figure 32. The Train song was the only unfamiliar song out of these songs. Rachel’s favorite song was the Nut song (93%). Her eyes usually lit up when she chose and heard the Nut song. She was extremely attentive when listening to it and vocalized more than usual after the song was sung.

The least selected songs were Elephant, Turtle, Fish, Rabbit and Tree, and the percentages of the number of times Rachel chose these songs are illustrated in Figure 32. Out of these, the Rabbit song was the only unfamiliar song.

Rachel’s least preferred song was the Elephant song (0%). She never chose it intentionally, but the song was sung to her during one time when her choice was prompted. She was also familiar with the song. At that single hearing she did not seem to take and interest in the song and closed her eyes a few times.

Figure 32. Rachel’s song selection.
**Most selected** songs were Nut, Spider, Bee, Monkey, and Chick, and the percentages of the number of times Tali chose these songs is illustrated in Figure 33. Out of the five, the Chick song was the only unfamiliar song.

Tali’s favorite song was the Nut song (100%). She would move her body, rock and move her head during the whole song. She also vocalized more than the ordinary after the song was sung to her. Typically, she would stop the rocking and head movement when the song ended, look at the researcher and begin to vocalize. For both Rachel and Tali this song was energizing and encouraged and stimulated vocalization.

**The least selected** songs were: Elephant, Rabbit, Crocodile, Tree and Turtle, and the percentages of the number of times Tali chose these songs is illustrated in Figure 33. Out of these, the Rabbit and the Crocodile were unfamiliar songs. The Elephant song was never selected.

Tali’s least favorite song was the Elephant song (0%). Similarly to Rachel, Tali never chose the song intentionally, but heard it during a prompting trial. She was also familiar with the song. And while hearing it she barely moved her body, her head (as contrary to her usual manner while listening to songs) and her facial expression was a serious one.
TALIA

The most selected songs were: Bee, Nut, Monkey, Spider and Butterfly, and the percentages of the number of times Talia chose these songs is illustrated in Figure 34. All five were familiar songs.

Talia’s most preferred song was the Bee song (86%). Talia typically smiled during this song, which changed into a wide smile during the vocal play in the song. Talia’s mood had been down for a few months and as a result her motivation was down. She avoided choosing songs during group music therapy and may have needed the individual attention that was available to her during the study. Such intimate former knowledge of the participant strengthens the notion that a combination of researcher/clinician can yield better understanding of the data and allows for a more in depth analysis.
Least selected songs were: Fish, Hands, Rabbit, Elephant and Turtle, and the percentages of the number of times Talia chose these songs is illustrated in Figure 34. Out of these, the Hands and the Rabbit songs were unfamiliar songs. The Fish song was never selected.

**Figure 3. Talia’s song selection.**
### 3.4.2 Song selections and preferences in the group

The results reveal that three of the participants had their individual preferred song. Aviv preferred the Train song, Hilla the Monkey song and Meirav the Spider song. Two participants shared the same preferred song: Elisheva and Talia’s most preferred song was the Bee song. Two more participants shared the same preferred song: Rachel and Tali’s most preferred song was the Nut song. All of their most preferred songs: Nut, Monkey, Spider, Bee and Train songs were the overall five most selected songs.

In the least preferred song category, three of the participants (Aviv, Elisheva and Meirav) shared the Rabbit as the least preferred song. Two participants (Hilla and Talia) shared the Fish song as their least preferred song and two other participants (Rachel and Tali) shared the Elephant song as their least preferred song. There was less disagreement as to the least preferred songs. The Fish, Rabbit and the Elephant songs were the three overall least preferred songs on the list of songs.

Only one song out of the five most preferred songs, the Train song, was unfamiliar to the participants, and was the fifth song on the list. The four other songs (Nut, Monkey, Spider and Bee songs) were all familiar songs to the participants.

Three out of the five least preferred songs (Rabbit, Crocodile and Hands) were unfamiliar to the participants prior to the study. Two songs (Fish and Elephant) were familiar songs.

The small graphs **Figure 35** demonstrate that despite individual preferences within the participants, there was a resemblance of preferred and non-preferred songs between them.
Figure 35. Individual song selection

Rachel’s song selection

Elisheva’s song selection

Tali’s song selection

Hilla’s song selection

Meirav’s song selection

Talia’s song selection

Aviv’s song selection

Song selection of RS girls—most to least preferred songs
3.5 EMOTIONAL, COMMUNICATIVE AND PATHOLOGICAL BEHAVIORS

Introduction

As the majority of individuals with Rett syndrome are not verbal communicators, some information can be drawn from their emotional and physical behaviors. Analyzing different behaviors can assist the caregiver and therapist in understanding the child, thus providing her with better interaction.

The following emotional, communicative and pathological responses were monitored and analyzed. All data collected was sub-categorized into three sections; emotional, communicative and pathologically related behaviors.

1. The emotional behaviors were categorized as: Smile, laughter, frown, cry, moan, shout, and vocalization.

2. The communicative behaviors were categorized as: Body movement (rocking), leg movement, eye-contact look away eye shift closure of eyes, leave seat, walk towards exit door, return to seat, lean forward, lean back, push picture symbols away, lean hands on researcher, puts leg on researcher, and open/closure of mouth.

3. The pathological behaviors - can be categorized as Hand movement, teeth grinding, and hyperventilation. Change in facial color could fall into either pathological or emotional behavior, depending on whether it reflected raised arousal or raised anxiety respectively. For the purpose of this analysis, it will be placed under pathological.

In conjunction with Sigafoos’s analysis (1999; 2000) all behaviors can be defined as non-conventional forms of communication, and as a form of ‘Potential Communicative Act’.
The following emotional, communicative and pathological behaviors were derived from the baseline and intervention. These were observed during the middle period of the five most preferred songs (Nut, Monkey, Spider, Bee and Train) and five least preferred songs (Rabbit Elephant, Fish, Crocodile and Hands). Observation of the same songs took place during the baseline period. Each observation during baseline was carried out for three minutes during choice making. These were selected from the middle of the period of these specific songs during baseline.

The responses were analyzed in relation to the two different kinds of music (preferred vs. non-preferred) and when choice making occurred (during baseline) (see Tables 16-25).

The analysis does not provide the amount of occurrences of these behaviors during baseline and intervention but gives information whether these existed or not. The presence or absence of these behaviors may have some individual significance. One participant may stop the hand movement, then look at the researcher. This may be significant. Another participant may increase the tempo of hand movement to the music. This may indicate raised levels of excitement and enthusiasm. Some participants may lean forwards at the end of a song that could suggest for “more”. These types of behaviors were carefully observed and analyzed in an attempt to find consistency and in order to give meaning to them.
3.5.1 Emotional, communicative and pathological behaviors in individual cases

Tables 16–22 document the emotional, communicative and pathological behaviors for each participant in the study during the baseline and intervention conditions. These behaviors sampled as described above, were analyzed and further sub-categorized in terms of their positive, neutral or negative attributes and can also be seen in these tables. The categories represent a qualitative interpretation of emotional, communicative and pathological behaviors.

Table 16. Aviv’s responses - a comparison between baseline and intervention.

<table>
<thead>
<tr>
<th>Behaviors</th>
<th>Baseline (No music)</th>
<th>Intervention Non-preferred songs (With music)</th>
<th>Intervention Preferred songs (With music)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Facial expression</td>
<td>Frown, no smile</td>
<td>Frown, smile (on/off)</td>
<td>Smile, wide/big smile</td>
</tr>
<tr>
<td>Eye contact</td>
<td>Turn head away, eye shifts</td>
<td>Eye contact</td>
<td>Eye contact</td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Laugh/cry</td>
<td>Cry</td>
<td></td>
<td>Laughter</td>
</tr>
<tr>
<td>Body movement</td>
<td>Leave seat towards exit door</td>
<td>Rock</td>
<td>Rock, leave seat and return</td>
</tr>
<tr>
<td>Leg movement</td>
<td></td>
<td></td>
<td>Put leg on researcher</td>
</tr>
<tr>
<td>Hand movement</td>
<td></td>
<td></td>
<td>Put hand on researcher, tap on guitar</td>
</tr>
<tr>
<td>Lean forward end of song</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hand movement</td>
<td>Low level</td>
<td>High level</td>
<td>High level</td>
</tr>
</tbody>
</table>

Index: Neutral | Negative | Slightly positive | Positive
Table 17. Elisheva’s responses - a comparison between baseline and intervention.

<table>
<thead>
<tr>
<th>Behaviors</th>
<th>Baseline (No music)</th>
<th>Intervention</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>Non-preferred songs (With music)</td>
</tr>
<tr>
<td>Facial expression</td>
<td>Frown, no smile</td>
<td>Frown, smile (on/off)</td>
</tr>
<tr>
<td>Eye contact</td>
<td>Turn head away, eye shifts</td>
<td>Eye contact</td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Laugh/cry</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td>Rock</td>
<td>Lean forward, Rock</td>
</tr>
<tr>
<td>Leg movement</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Look away end of song</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Push symbols away</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Breath holding</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Open/close mouth</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hand movement</td>
<td>Low level</td>
<td>High level</td>
</tr>
</tbody>
</table>

Index: Neutral | Negative | Slightly positive | Positive
Table 18. Hilla’s responses - a comparison between baseline and intervention.

<table>
<thead>
<tr>
<th>Behaviors</th>
<th>Baseline</th>
<th>Intervention</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>(No music)</td>
<td>Non-preferred songs</td>
</tr>
<tr>
<td></td>
<td></td>
<td>(With music)</td>
</tr>
<tr>
<td>Facial expression</td>
<td>Frown, no smiles</td>
<td>Frown, smile (on/off)</td>
</tr>
<tr>
<td>Eye contact</td>
<td>Stare</td>
<td>Eye contact</td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Laugh/cry</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td>Lean back</td>
<td>Rock</td>
</tr>
<tr>
<td>Push symbols away</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Look away at end of song</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Open/close mouth</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Breath holding</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hand movement</td>
<td>Low level</td>
<td>Low level</td>
</tr>
</tbody>
</table>

**Index:** Neutral    Negative    Slightly positive    Positive
Table 19. Meirav’s responses - a comparison between baseline and intervention.

<table>
<thead>
<tr>
<th></th>
<th>Baseline (No music)</th>
<th>Intervention Non-preferred songs (With music)</th>
<th>Intervention Preferred songs (With music)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Facial expression</td>
<td>Frown, no smile</td>
<td>Frown, smile (on/off)</td>
<td>Smile, wide/big smile</td>
</tr>
<tr>
<td>Eye contact</td>
<td>Look away</td>
<td>Eye contact</td>
<td>Eye contact</td>
</tr>
<tr>
<td>Vocalization</td>
<td>Shout</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Laugh/cry</td>
<td>Cry</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td>Rock</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Leg movement</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Look away end of song</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hand movement</td>
<td>High level</td>
<td>Low level</td>
<td>High level</td>
</tr>
</tbody>
</table>

**Index:** Neutral | Negative | Slightly positive | Positive
Table 20. Rachel’s responses- a comparison between baseline and intervention.

<table>
<thead>
<tr>
<th>Behaviors</th>
<th>Baseline</th>
<th>Intervention</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>(No music)</td>
<td>Non-preferred songs (With music)</td>
</tr>
<tr>
<td>Facial expression</td>
<td>Frown, no smile</td>
<td>Frown, smile (on/off)</td>
</tr>
<tr>
<td>Eye contact</td>
<td>Close eyes</td>
<td>Eye contact</td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Laugh/cry</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td></td>
<td>Rock</td>
</tr>
<tr>
<td>Open/close mouth</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Look away at end of song</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hand movement</td>
<td>Low level</td>
<td>Low level</td>
</tr>
</tbody>
</table>

**Index:** Neutral Negative Slightly positive Positive
Table 21. Tali’s responses - a comparison between baseline and intervention.

<table>
<thead>
<tr>
<th>Behaviors</th>
<th>Baseline (No music)</th>
<th>Intervention Non-preferred songs (With music)</th>
<th>Intervention Preferred songs (With music)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Facial expression</td>
<td>Frown, no smile</td>
<td>Frown, smile (on/off)</td>
<td>Smile, wide/big smile</td>
</tr>
<tr>
<td>Eye contact</td>
<td>Close eyes</td>
<td>Eye contact</td>
<td>Eye contact</td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td></td>
<td>Shout</td>
</tr>
<tr>
<td>Head movement</td>
<td></td>
<td>Rock</td>
<td>Lean forward, Rock</td>
</tr>
<tr>
<td>Body movement</td>
<td></td>
<td>Rock</td>
<td>Lean forward, Rock</td>
</tr>
<tr>
<td>Hand movement</td>
<td>High level</td>
<td>Low level</td>
<td>Low level</td>
</tr>
</tbody>
</table>

Index: Neutral, Negative, Slightly positive, Positive
Table 22. Talia’s responses - a comparison between baseline and intervention.

<table>
<thead>
<tr>
<th>Behaviors</th>
<th>Baseline (No music)</th>
<th>Intervention Non-preferred songs (With music)</th>
<th>Intervention Preferred songs (With music)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Facial expression</td>
<td>Frown, no smile</td>
<td>Frown, smile (on/off)</td>
<td>Smile, wide/big smile</td>
</tr>
<tr>
<td>Eye contact</td>
<td>Close eyes, Look away</td>
<td>Eye contact</td>
<td>Eye contact</td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td>Moan</td>
<td></td>
</tr>
<tr>
<td>Teeth grinding</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td></td>
<td>Lean forward</td>
<td></td>
</tr>
<tr>
<td>Change in facial color</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hand movement</td>
<td>Low level</td>
<td>Low level</td>
<td>High level</td>
</tr>
</tbody>
</table>

Index: Neutral, Negative, Slightly positive, Positive
What can be seen in the above tables are both a wide variety in styles of emotional, communicative and pathological response, and also a range of positive to negative degrees of intensity. This part of the analysis relies mainly on a qualitative interpretation of the behaviors, which is informed by the fact that the researcher is also the therapist. The researcher has both prior knowledge of the participants, and extra-music therapy knowledge (by which one means the knowledge of these participants in situations other than music therapy). While it was not intended in this research to make any comparative evaluation of response in different situations, the categorization of these types of responses (emotional, communicative and pathological) is informed by an understanding of these participants within a wider institutional context.

3.5.2 Analysis of one participants emotional responses to one song over time

The Train song (Figure 36) (an unfamiliar song when initially presented and a preferred one) can illustrate the participant’s emotional and communicative responses to the music, and how these responses developed and changed over a number of presentations over several weeks. This will illustrate the process of emotional and communicative responsiveness.

In the Train song it is worth noticing the process of the one participant (Aviv) emotional response. There is a buildup in Aviv’s emotional and communicative responses during different events in the music, followed by a decline. It is like an exciting stimulus, which becomes familiar and then the responses fades away and becomes habituated. This example shows the number of times (14 times) this song was chosen and sung to the participant.
It reveals a consistent progression of her response. In the third measure of the song, Aviv looked up during the first few times she heard the sounds "toot toot" (notes a a – A A). Later into hearing the song she smiled during the sound "toot toot" and as the song became familiar one she burst into laughter exactly at the same place in the music. The emotional response was gradually enhanced with smiles and laughter. As the sessions progressed and this song had been heard several times, Aviv smiled widely in same place, but stopped laughing. During the last sessions she made a smaller smile and at the end there were no smiles and Aviv left her seat and walked towards the door (a behavior she had previously done when she heard the song for the first time).

Aviv’s response to the musical events was on the right timing and at times, she even anticipated the sound that was about to be heard.

This type of in emotional and communicative buildup and decline over time was very well demonstrated by all the participants, but was especially clear with Aviv in the Train song.
Figure 36. Train song - emotional and communicative buildup and decline over time.

<table>
<thead>
<tr>
<th>Date</th>
<th>1</th>
<th>2</th>
<th>3</th>
</tr>
</thead>
<tbody>
<tr>
<td>14/6/99</td>
<td>L</td>
<td></td>
<td></td>
</tr>
<tr>
<td>11/6/99</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>21/6/99</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1/7/99</td>
<td>L</td>
<td></td>
<td></td>
</tr>
<tr>
<td>5/8/99</td>
<td>L</td>
<td></td>
<td></td>
</tr>
<tr>
<td>15/9/99</td>
<td>BS</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Index:  
L - Laughter, Long laughter  
S - Smile  
BS – Big smile  
△ looks up at therapist  
1 – First chorus of the train song  
2,3 – repetitions of the train song
### 3.5.3 Emotional, communicative and pathological behaviors as a group

All responses were recorded for all girls in the three different selected areas observed: Baseline (no music), intervention with non-preferred songs and with most preferred songs (with music).

#### Table 23. All girls - Responses during baseline.

<table>
<thead>
<tr>
<th>Child</th>
<th>Aviv</th>
<th>Elisheva</th>
<th>Hilla</th>
<th>Meirav</th>
<th>Rachel</th>
<th>Tali</th>
<th>Talia</th>
</tr>
</thead>
<tbody>
<tr>
<td>Behaviors</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Eye contact</td>
<td>Turn head away, eye shifts</td>
<td>Turn head away, eye shifts</td>
<td>Stares at researcher</td>
<td>Look away</td>
<td>Close eyes</td>
<td>Close eyes</td>
<td>Close eyes, Look away</td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td>Shout</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cry/laugh</td>
<td></td>
<td>Cry</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td>Leave towards exit</td>
<td>Lean back</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hand movement</td>
<td>Push symbols away</td>
<td>Push symbols</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Leg movement</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Head movement</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Lean forward at end of song</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Breath holding</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Teeth grinding</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Open/closes mouth</td>
<td></td>
<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Change in facial color</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Stereotypic hand movement</td>
<td>Low level</td>
<td>Low level</td>
<td>Low level</td>
<td>High level</td>
<td>Low level</td>
<td>High level</td>
<td>Low level</td>
</tr>
</tbody>
</table>

**Index:** Neutral | Negative | Slightly positive | Positive
The purpose of the above tables was to group all emotional, communicative and pathological behaviors of all participants to see if any typical patterns of behavior emerged.

Table 23 shows that facial reactions, such as a frowning face, and lack of a smile, appear with all subjects in a situation that can be hypothesized as “not liked”. Other behaviors, extremely common in the population in disliked situations are lack of eye contact (86%), lack of vocalization or shouting (100%). Some girls showed different individual behaviors expressing their unease or rejection of the situation. Such behaviors were:

- Crying (2/7)
- Breath holding (2/7)
- Hand movements – pushing the symbols away (2/7)
- Body movement – leaving the chair and walking towards the exit, leaning backward (2/7)
- Teeth grinding (1/7)

Stereotypic hand movements show a certain direction as 5 out of the 7 (71%) girls participating in the present research showed a low level of hand mannerisms.
Table 24. All girls - Responses during intervention (non preferred songs).

<table>
<thead>
<tr>
<th>Child</th>
<th>Aviv</th>
<th>Elisheva</th>
<th>Hilla</th>
<th>Meirav</th>
<th>Rachel</th>
<th>Tali</th>
<th>Talia</th>
</tr>
</thead>
<tbody>
<tr>
<td>Behaviors</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Eye contact</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Moan</td>
</tr>
<tr>
<td>Cry/laugh</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td>Rock</td>
<td>Rock</td>
<td>Rock</td>
<td>Rock</td>
<td>Rock</td>
<td>Rock</td>
<td></td>
</tr>
<tr>
<td>Hand movement</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Leg movement</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Head movement</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Lean forward at end of song</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Breath holding</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Teeth grinding</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Open/closes mouth</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Change in facial color</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Stereotypic hand movement</td>
<td>High level</td>
<td>High level</td>
<td>Low level</td>
<td>Low level</td>
<td>Low level</td>
<td>Low level</td>
<td>High level</td>
</tr>
</tbody>
</table>

Index: Neutral | Negative | Slightly positive | Positive

Table 24 reveals that the most evident changes in behaviors detected when introducing the songs to the participants were as follows:

- Eye contact by all girls (100%).
- Smiles detected on and off on all faces, although the constant expression tended to be a frown (100%).
• Lack of vocalization or moaning (100%).
• The appearance of rocking as a response to the introduction of music in 6 out of 7 participants (86%).
• The overall picture of a decreased level of activity in all parts of the body.

Stereotypic hand movements changed from baseline (mostly high level of hand mannerisms - 5/7) to a mixed appearance. Four out of seven of the participants (57%) showed a low level of activity, whereas 3/7 (43%) percent showed high levels of activity.

Table 25 represents a situation that can be defined as a preferred or “liked” situation.

In general, activity level seems much higher when compared with the activity level observed when the participants were exposed to non-preferred music. Activities characteristic of participants when listening to their choice of songs were:

• Facial expression – wide, big, and continuous smiles were detected with all (100%) participants.
• Eye contact was observed with all (100%) participants.
• Vocalization was apparent in all (100%) cases.
• Different body movements (such as: Rocking, leaving seat and returning, leaning forward) was evident with all (100%) participants.
• Body movement (head movement, hand movements, and leg movements) was recorded in 4 out of 7 participants (57%).

Laughing was a dominant behavior seen in 5 out of 7 (71%) girls. Such behavior was not recorded in other situations.
<table>
<thead>
<tr>
<th>Child</th>
<th>Aviv</th>
<th>Elisheva</th>
<th>Hilla</th>
<th>Meirav</th>
<th>Rachel</th>
<th>Tali</th>
<th>Talia</th>
</tr>
</thead>
<tbody>
<tr>
<td>Behaviors</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Facial expression</td>
<td>Smile, wide smile</td>
<td>Smile, wide smile</td>
<td>Smile, wide smile</td>
<td>Smile, wide smile</td>
<td>Smile, wide smile</td>
<td>Smile, wide smile</td>
<td>Smile, wide smile</td>
</tr>
<tr>
<td>Eye contact</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vocalization</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Shout</td>
<td></td>
</tr>
<tr>
<td>Cry/laugh</td>
<td>Laugh</td>
<td>Laugh</td>
<td>Laugh</td>
<td>Laugh</td>
<td>Laugh</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td>Rock, leave seat and return, lean forward</td>
<td>Rock, lean forward</td>
<td>Rock, lean forward</td>
<td>Rock</td>
<td>Rock, Lean forward</td>
<td>Rock, lean forward</td>
<td>Lean forward</td>
</tr>
<tr>
<td>Hand movement</td>
<td>Tap on guitar</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Leg movement</td>
<td>Put leg on researcher</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Head movement</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Breath holding</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Teeth grinding</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Open/closes mouth</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Change in facial color</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Look away at end of song</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Stereotypic hand movement</td>
<td>High level</td>
<td>High level</td>
<td>High level</td>
<td>High level</td>
<td>High level</td>
<td>Low level</td>
<td>High level</td>
</tr>
</tbody>
</table>

**Index:** Neutral  | Negative  | Slightly positive | Positive
Other behaviors were apparent only when preferred songs were played were:

- Looking away at the end of each song (3/7)
- Opening and closing mouth as the song was played (3/7)
- Change in facial color (1/7).
- Stereotypic hand movements appeared for most girls (6/7; 86%), at a high level.
3.5.4 Emotional and communicative behaviors – group analysis of one song

The communicative and emotional behaviors can be observed in the music during all songs, but will be demonstrated during one preferred song (the Bee song).

Figure 37. Bee song – Group emotional and communicative behaviors.

<p>| | | | |</p>
<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Aviv</td>
<td>S</td>
<td>BS</td>
<td>BS</td>
</tr>
<tr>
<td>Meirav</td>
<td>BS</td>
<td>L</td>
<td>L</td>
</tr>
<tr>
<td>Tali</td>
<td>S</td>
<td>BS</td>
<td>BS</td>
</tr>
<tr>
<td>Elishva</td>
<td>S</td>
<td>BS</td>
<td>BS</td>
</tr>
<tr>
<td>Rahel</td>
<td>BS</td>
<td>S</td>
<td>S</td>
</tr>
<tr>
<td>Hilla</td>
<td>S</td>
<td>S</td>
<td>S</td>
</tr>
<tr>
<td>Talia: Looks up</td>
<td>S</td>
<td>S</td>
<td>S</td>
</tr>
</tbody>
</table>
The Bee song was one of the five most preferred songs. The score above show all seven participants (during the second part of the song) indicating some emotional responses on the exact same place in the music (measures 7-12). It occurred when there was an interesting vocal play such as “oops” and “bzz”.

The ‘S’ stands for a Smile, the ‘BS’ stands for a ‘Big Smile” and the ‘L’ stands for a Laugh.

Each participant responded during these places, but each one responded differently. The first time the ‘oops’ sound was heard, the participants either smiled, made a big smile or did not smile at all. The second and third time the ‘oops’ sound was heard Aviv, Meirav, Tali, Elisheva and Talia’s emotional responses enhanced. The response was typically from smile to big smile, from big smile to laughter or from no smile to a smile. Hilla continued to smile during the ‘oops’ sound and Rachel’s emotional response decreased from big smile to a smile.

The reason for the enhancement in emotional response can be due to the ‘bzz’ sound that was heard just before the second and third time the ‘oops’ sound appeared. This may have enhanced tension for the anticipated ‘oops’ sound. All participants demonstrated positive emotional response during the second and third time the ‘oops’ sound was heard.
Summary

The results of this analysis of emotional, communicative and pathological behavior in all the participants can be generally summarized as follows:

**Baseline:**
Frowning faces with no smiles, crying, no vocalization, no eye contact, and low level of hand movement are characteristic during the baseline period.

**Intervention:**
Non-preferred songs - The appearance of eye contact with sparse smiles together with a typically serious face, a reduced level of bodily activity and a small rise in stereotypic hand movement mark the change from no music at all to listening to non-preferred music.

Preferred songs - Continuous eye contact, accompanied by wide big smiles and laughing, vocalization, an overall high level of bodily activity, and a high level of hand movement was apparent from the moment a preferred song was played for the participants in the present study.

Conclusion

This concludes the presentation of the results from this study, which has been documented in order to present the participants responses and the degree of intentional choice making as well as other aspects. The results in this chapter have included a level of analysis of their response time, their learning processes in particular considering their ability to learn over time, and also their song preferences and their emotional, communicative and pathological behaviors.
These results were achieved in response to the specific research questions were outlined in the literature review chapter in order to determine whether the participants were able to make choices, how they made choices and how that developed over time. The evidence from the analysis also revealed aspects of their behavior in the support of their choice making.

An area of analysis that was undertaken in order to consider the rational behind their choice of songs was the songs themselves, and also the familiarity to the songs together with the checking whether there is any influence relating to the colors used in the symbols that were presented to the participants when choosing.

The analysis of the songs expanded to quite a significant degree as it revealed some characteristics and musical elements that might have influenced either preference or non-preference of songs. Therefore, rather than include this material in the results chapter (related specifically to the research questions that were asked), it was decided to document it in a separate chapter together with the discussion of the implication of those specific results from the analysis of the musical material of the songs. The musical analysis results and discussion of the analysis will follow this chapter.
CHAPTER FOUR
SONG ANALYSIS
RESULTS with DISCUSSION

INTRODUCTION

The 18 songs used in this research were structurally analyzed to determine what musical features they may contain. The following features were included: familiarity, tempo, meter, key signature, tempo variability such as: fermatas, pauses, accelerando, ritardando, upbeat, syncopation.

Other features such as rhythmical patterns, melodic motifs, dynamics and vocal play were analyzed.

Table 26. List of musical features.

<table>
<thead>
<tr>
<th>Song</th>
<th>Tempo</th>
<th>Key</th>
<th>Meter</th>
<th>Upbeat</th>
<th>Fermata/pauses</th>
<th>Ritardando</th>
<th>Accelerando</th>
<th>Vocal play</th>
</tr>
</thead>
<tbody>
<tr>
<td>Nut</td>
<td>144</td>
<td>A dur</td>
<td>4/4</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
</tr>
<tr>
<td>Monkey</td>
<td>184</td>
<td>D dur</td>
<td>2/4</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
</tr>
<tr>
<td>Spider</td>
<td>104</td>
<td>A dur</td>
<td>6/8</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
</tr>
<tr>
<td>Bee</td>
<td>160</td>
<td>D dur</td>
<td>4/4</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
</tr>
<tr>
<td>Train</td>
<td>132</td>
<td>A dur</td>
<td>6/8</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
</tr>
<tr>
<td>Farm</td>
<td>132</td>
<td>C dur</td>
<td>4/4</td>
<td>V</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bird</td>
<td>152</td>
<td>D dur</td>
<td>4/4</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
</tr>
<tr>
<td>Chick</td>
<td>120</td>
<td>D dur</td>
<td>3/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Butterfly</td>
<td>100</td>
<td>A dur</td>
<td>4/4</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
<td>V</td>
</tr>
<tr>
<td>Frog</td>
<td>112</td>
<td>A dur</td>
<td>2/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Star</td>
<td>96</td>
<td>D dur</td>
<td>4/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Tree</td>
<td>84</td>
<td>A moll</td>
<td>4/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Turtle</td>
<td>84</td>
<td>A dur</td>
<td>4/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Hands</td>
<td>72</td>
<td>D dur</td>
<td>4/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Crocodile</td>
<td>72</td>
<td>Chant</td>
<td>4/4</td>
<td>V</td>
<td>V</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Fish</td>
<td>92</td>
<td>A dur</td>
<td>4/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Elephant</td>
<td>90</td>
<td>A dur</td>
<td>4/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Rabbit</td>
<td>92</td>
<td>A dur</td>
<td>4/4</td>
<td>V</td>
<td></td>
<td>V</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
In addition, the colors of the picture symbols used in the choice making procedure were considered to determine any influence they may have had. There will also be a short discussion at the end of this chapter regarding the performance of the songs in this study.

Stemming from the list in Table 26 one can begin to make inference as to what determined most or least preferred songs. The five most preferred songs (Nut, Monkey, Spider, Bee and Train) and the five least preferred songs (Rabbit, Elephant, Fish, Crocodile and Hands) will be analyzed. Scores of all songs can be found in Appendix IX and are listed according to most to least preferred songs. All songs in that order can be heard in the CD attached to the thesis. Although only five most and five least preferred songs will be analyzed, it is interesting to note that the closer the rest of the songs are to the top or the bottom of the preference list, the more in common they have with the five most and least preferred songs.

4.1 FAMILIARITY

The participants were familiar with twelve out of the eighteen songs. These were randomly categorized into sets of four familiar and two unfamiliar songs, a total of three sets. The participants chose out of these sets and had many opportunities in making choices between familiar and unfamiliar songs. Four out of the five most preferred songs were familiar while only one was unfamiliar. This applies to the whole group. Looking at the unfamiliar songs within each individual case, some girls chose unfamiliar songs in the most preferred song list, while others did not. Aviv, Meirav and Rachel chose the Train song and Elisheva and Tali chose the Chick song. Talia and Hilla stayed with familiar songs when choosing the top five songs. These five girls only chose one unfamiliar song out of all preferred songs. Although the
Train song was an unfamiliar song, much of its content (rhythm and melody) was similar to the Spider song (a familiar song).

Table 27. Familiar/non-familiar songs vs. Most/least preferred songs – True results.

<table>
<thead>
<tr>
<th></th>
<th>5 most preferred songs (X5=35)</th>
<th>5 least preferred songs (X5=35)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Familiar</td>
<td>30</td>
<td>13</td>
</tr>
<tr>
<td>Non-familiar</td>
<td>5</td>
<td>22</td>
</tr>
</tbody>
</table>

When looking at the above-presented table one might observe those familiar songs are strongly presented at the preferred songs group, while non-familiar songs are prominent in the least preferred songs group. An unbiased preference (not effected by the familiarity) will leave songs (familiar or unfamiliar) at the same ratio (12:6 or 2:1 - as their proportion among all song used in the present study) in both groups. Such a graph will present the following figures:

Table 28. Familiar/non-familiar songs vs. Most/Least preferred songs – expected results.

<table>
<thead>
<tr>
<th></th>
<th>5 most preferred songs (X5=35)</th>
<th>5 least preferred songs (X5=35)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Familiar</td>
<td>23.4</td>
<td>23.4</td>
</tr>
<tr>
<td>Non-familiar</td>
<td>11.6</td>
<td>11.6</td>
</tr>
</tbody>
</table>

A chi-square test reveals the fact that the difference between the expected un-biased results (Table 28) and the true findings (Table 27) is statistically significant (P<0.00001), presenting the fact that familiarity has an effect on song preference of children with Rett syndrome.

Two out of the five least preferred songs were songs familiar to the group. The remaining three, were unfamiliar songs. There are more unfamiliar songs in the least preferred songs when viewing individual cases.
Aviv and Hilla had four unfamiliar songs in the least preferred songs, which was 66% out of the unfamiliar songs presented in the study. Meirav and Elisheva had three unfamiliar songs in the least preferred (50% of all unfamiliar songs in the study) and all the remaining girls had two unfamiliar songs (33% of all unfamiliar songs in the study).

Some reports claim that unfamiliar songs might stimulate interest by the listener (Bruscia, 1987), although this does not correspond to the finding of the present study. It might be due to other factors such as musical elements discussed elsewhere, or it might be due to the fact that these songs had to be sung more often in order for reactions to them to have become stabilized.

One can infer from these findings that although two unfamiliar songs were chosen by five of the participants, familiarity is a vital factor when choosing songs by females with Rett syndrome. These findings might be explained in light of the fact that familiarity creates comfort and security (Bruscia, 1987). The girls preferred familiar songs, ones they were comfortable with and were known to them. The girls could anticipate and look forwards to these songs. Familiarity may have provided a secure feeling, without too many unexpected changes. This finding corresponds with the finding of Iacono, Carter & Hook (1998) who tried to identify intentional communication in children with severe and multiple disabilities and found that familiarity involving listening to music resulted in more communication within their participants. Other studies have shown that the use of more familiar material while teaching may promote greater learning (Gfeller, 1983; McGuire, 2001).

Further findings by researchers and clinicians support the present one that girls with Rett syndrome who selected familiar songs became more animated and generated greater communication and responsiveness when songs are heard (Braithwaite & Sigafoos, 1998; Elefant and Lotan, 1998; Hadsell and Coleman, 1988; Holdsworth,
1999; Woodyatt & Ozanne, 1992, 1994). A diminished responsiveness was detected by Holdsworth, 1999 with unfamiliar music as in the present study.

Unfamiliarity is very likely to be a contributory factor to song preference. Two explanations can be offered to account for the limited number of unfamiliar songs in the most preferred songs and on the other hand, the clear predominance of such songs in the least preferred songs. The first explanation could be due to the novelty of the songs. The participants seemed to prefer songs with which they were familiar. The second explanation could be that the least preferred songs (including the unfamiliar songs) were musically un-satisfying. This will be discussed at length further on. Although all of the participants chose unfamiliar songs during the study, they only returned to the ones they seemed to like, and never returned back to the other unfamiliar songs.

4.2 MUSICAL FEATURES

The full scores of all 18 songs can be found in Appendix IX

4.2.1 Tempo

There was a dramatic difference in tempo between the five most and the five least preferred songs. The mean tempi of the five most preferred songs was 144.8 beats per minute, while the mean tempi of the five least preferred songs was 83.6 beats per minute.

All songs in the most preferred category in individual cases were at least or more than 100 beats per minute (Figure 38).
The tempo seems to have a big effect on whether or not the participants preferred the songs, as they were typically drawn to music with faster tempi. This reinforces other findings that girls with Rett syndrome appear to prefer energetic music (Holdsworth, 1999). One suggestion for this is that the fast tempo as the external stimuli (the songs) may be in synchrony with these girls’ internal level of arousal/rhythm, which is observed and reported to be at a constantly high level due to over activation of the sympathetic nervous system (Julu, 2000). Another suggestion for explaining the preference of faster tempi may be that since girls with Rett syndrome are limited in their initiation of purposeful activity, they may respond enthusiastically when being activated ‘emotionally’. Fast tempi in music brings vitality and energy into their lives while slower tempi may provoke their potential passivity, since the slow tempo causes the listener to lose sense of rhythm or musical motion (Wittmann & Poppel, 1999). Another possibility for preferring faster tempi might be connected to the fact that slow music has been reported to increase anxiety levels in individuals with Rett syndrome (Mount, et. al., 2001). Better yet, a very simple explanation for fast tempi preference by girls with Rett syndrome is due to their “normal” attitudes towards
music. Their song preference is similar to that of professional musicians/music therapists and research has shown that children in the same age group as this research population prefer music with a fast rather than a slow tempo (LeBlanc, 1981; LeBlanc & Cote, 1983; LeBlanc & McCrary, 1983; Sims, 1987).

Choosing songs during intervention and hearing them was a rewarding and motivating experience and as a result the overall energy level during the research sessions was at a faster pace. This could have contributed and determined the participants’ choice in songs with faster tempo.

4.2.2 Meter

The meter in the preferred songs were 2/4 or 6/8, while the meter for the least preferred songs were 4/4. The metric organization is the differentiation of pulse into accented and unaccented beats (Meyer, 1956). The feeling of more accents as part of the most preferred songs could have contributed to the faster feeling, as well as to their level of excitement.

4.2.3 Key signature

Looking at the influence of key, one can see that both the most preferred songs, and also the least preferred songs were in the key of either D major, A major or C major. No conclusions regarding the influence of different keys can therefore be drawn in terms of song preference.
4.3 TEMPO AND RHYTHMIC VARIABILITY

4.3.1 Ritardando (slowing down)

All of the favorite songs had ritardando, while only one out of the least preferred songs had ritardando. Events in the songs where the music slowed down can be heard before accelerations, before a new phrase, towards the end of a verse, towards the end of the song and before an interesting vocal play or high pitch. The ritardando may give an illusion of slowing down/calming down with the anticipation that something is about to happen. It seems that the ritardando is an important feature when determining preference or when composing songs for this population.

4.3.2 Accelerando (moving forwards)

Accelerando featured in all preferred songs, while only one of the least preferred songs had accelerando. Accelerating in the music gives new energy to the tempo. It builds up tension by not knowing where it is going, but with a feeling that it will reach an end. Accelerando elicits excitement in the music. The results suggest that an accelerando is an important feature when determining preference or when composing songs for this population.

4.3.3 Fermata (holding note) and Pauses

All preferred songs and three of the least preferred songs had fermatas and pauses. The pauses in the two least preferred songs are found at the end of the song, while in
preferred songs they appear several times during the songs. Fermata and pauses build up tension and excitement and invite response. Fermata may be a cause for emotional arousal as the listener’s expectation is temporarily held (Meyer, 1956) and it can provoke playfulness and humoristic teasing (Burford & Trevarthen, 1997). All of these seem to be important factors when choosing preferred songs or when composing songs for this population.

4.3.4 Upbeat introductions and Syncopation

All favorite songs have upbeat (pick-up) before the first measurement. They all have syncopations throughout the songs. One of the least preferred songs (Rabbit song) begins with an up beat and also has syncopation. But these are not as strong as in the other songs, and they are almost not heard.

There are different ways in which a rhythm can be changed in the course of a tune. In each case it is clear that expectancies and violation of expectancies are playing a major part in the promotion of emotional reaction to the music (Meyer, 1956; Sloboda, 1991).

Syncopation evokes response and has attributes that promote rhythmical tension. It adds vitality and excitement to the music and moves it forward. Because these girls enjoy fast, stimulating music for their songs, it can be argued that syncopation, or jazzy rhythms, will attract them significantly more than a regular meter and rhythm. Therefore an upbeat introductions, and syncopated patterns in the music are likely to be important features when determining preference or when composing songs for this population.
4.3.5 Rhythmic grouping

All favorite songs have rhythmic groupings of Iambic, Anapestic and Trochaic, while the non-preferred songs have mostly quarter note motions. The rhythmical patterns in the most preferred songs have interesting rhythms with rhythmic energy. The Bee song has a pattern of eighth dotted and sixteenth notes. The pattern is symmetrical throughout the song. The Spider song has an eighth, quarter, eighth, etc. rhythmical pattern that repeats itself. The Monkey song has sixteenth note rhythmical patterns and also contains rhythms such as dotted sixteenth and thirty-second note values. The Train song has rhythmical patterns of - eighth, quarter, eighth, quarter note, being constantly repeated. The Nut song has rhythmical patterns with lots of syncopations.

The least preferred songs have rhythmical patterns that remain rather static and have almost no rhythmical development. There are almost no rhythmical surprises and barely any changes in the rhythmical pattern to break the monotony. All least preferred songs are built on quarter or eighth note patterns that repeat themselves from the beginning to the end of the song. An example, the Rabbit song is built on rhythmical patterns of dotted eighth and sixteenth notes consecutively four times and then a quarter note throughout the entire song.

The rhythmical patterns in the songs add rhythmic tension and elicit response since early events in a musical sequence generate expectancies about events that are going to occur (Fraisse, 1982; Martin, 1972; Meyer, 1956). The Nut song has interesting rhythmical patterns with lots of syncopations. This gives a feeling of a ‘swing type’ motion.

Repetitive rhythms with unbound freedom (similar to the repetitive rhythms found in the songs in this study), was reported by Loewy (1995) to encourage and foster the
child’s inner rhythm to emerge and stabilize. She further states that the child’s physiological inner body rhythm can become activated by the reinforcement of the repetitive rhythmical patterns in the songs. In the least preferred songs, repetitive rhythmical patterns may cause boredom and tiredness in the listener where there are no other musical characteristics such as syncopation, fermata, tempo or rhythmic variability to attract or sustain interest. They may also elicit agitation because of the perpetual rhythmical patterns. Such agitation or uneasy feelings might be expressed in actions such as teeth grinding or leaving one’s seat and heading for the exit door, as was noted and will be discussed in length later.

4.4 MELODIC MOTIF

“Melody is an important aspect of musical expression, it is related to inner experiences and memory, and can function as an intimate companion along the various paths of one’s life” (Aldridge, 1999, p.142). Melodic contour is an important feature in melody perception from early infancy (Umemoto, 1997) and therefore it seems only natural that melodic motifs will have an important impact on the girls when preference is determined. Melody is an important form of expression (Aldridge, 1999) and girls with Rett syndrome need to express their emotions. As they don’t have any verbal means to express these emotions they can do so by actively listening to the melodies in the songs. The melodic developments are more interesting and with more variability in the most preferred songs when compared to the least preferred songs. The girls in the study seem to be sensitive and attend to these developments.
**Nut Song:** The Nut song has a clear motif, which repeats itself. The first interval is a descending minor third. There is a pentatonic element in the beginning of the song because of the minor third and the ascending fourth.

There are not as many intervals of seconds in this song when compared to other songs. There is a surprising change in measure eight, which gives the impression that the song has come to an end. The note E repeats itself many times, almost like a drone. This idea of a note repeating itself, provides a point of reference and security, and on the other hand the melody needs freedom. The melody is free and gives a ‘swing-like’ feeling.

**Monkey Song:** The Monkey song begins on the dominant with an upbeat, and the same note is repeated three times. This is followed by a large melodic jump of a major sixth, which contributes to the tension and it has certain mysteriousness in this interval and brings about a reaction. The sixth is resolved into a fifth. The dominant (measure 5) resolves itself to the tonic. Towards the end of the song there is a step-by-step decline till it reaches the tonic. There are many different intervals in this song: ascending sixth and ascending and descending fifth, descending fourth and an ascending fourth after the fermata, causing a surprise. Some of the large intervals break the melodic pattern, which makes it surprising and exciting. The song is uplifting because of its melodic mood. Some of the intrinsic characters in the melody enhance this feeling. There is movement in the melody and a freedom similar to the freedom felt in the Nut song.

**Spider Song:** The Spider song has many of the same melodic contours as the Train song but in this song there are many more pauses. These pauses help in emphasizing
the phrasing in the song. The interval range in the song is octave: middle E to high E. The melodic patterns contain three notes. The notes are based on a triad chord. The first motive is equal to the second, only a third tone higher. Measures 1 and 2 have a 3 step ascending notes, and thereafter measures 2 and 3 have a 3 step descending notes.

**Bee song:** The Bee song is based on a triad using the pentatonic scale. The melody is light and flowing. There is an interval of a fourth in the beginning of the song, then the melody moves upwards in steps. The fourth brings tension from the beginning. The first phrase is repeated identically in measure 5, then in measure 7. After that, there is a repeated chorus. Measure 8 has a glissando, ritardando and a surprise occurs (high A), which is the highest note in the song. The song is melodically symmetrical and with homogeneous phrase lengths.

The girls typically began smiling immediately when hearing the two first notes. The song has very symmetric phrasings with an open phrase then a closing leading to balance. This can lead to security and trust, which seems to be important to the girls.

**Train Song:** The Train song begins with a melodic motif of three notes that repeat on the chord notes. There is a descending fifth into the tonic and then back to the fifth. After that there is an ascending melodic direction of steps. Then a melodic development occurs and reaches a peak on a leap of an octave. There is a melodic tension throughout this song.

The five least preferred songs are very different when analyzing their melodic motifs.

**Rabbit Song:** The melody line of the Rabbit song is repetitive and within a limited range of notes. The intervals are mostly descending in half or whole steps or stand
on the same note. This can evoke a feeling of being pulled down as well as becoming heavy. The largest interval in the song is an ascending fourth, which comes up only twice during the song. The melody is predictable, repetitive and stays mostly the same with no melodic shades or changes. This can easily elicit boredom.

**Elephant song:** The Elephant song is made out of two phrases that repeat themselves. The whole song is based on descending steps, similarly to the Rabbit song. Twice, there is a large skip of an octave from middle A to an octave higher A only to begin the ascending notes of seconds. This song evokes feeling of being pulled down emotionally. The melodic patterns have no changes, its quality stay the same and is extremely predictable.

**Fish Song:** The fish song has melodic motif that repeats itself without a development in the melody. The song opens with a triad on the tonic. The first two notes repeat themselves twice and reach the dominant (first part of the phrase). Once on the dominant, the notes go down in steps to the tonic (second part of the phrase). This second part of the phrase repeats itself three times, beginning always on the same note. The continuation of the song repeats just as the first and second part of the phrase. The symmetry in the motive is off balance because of the ending and there are no pauses. There is no indication of breaking of the monotony. The melody gives a never-ending recitative like feeling. The ending of the song has an ascending motif that reaches the low E. It begins with a descending octave (middle E – low E), minor seventh (middle D - low E), major sixth (middle C# - low E), major fifth (lower B – lower E) and ends with a big skip to a middle A, brings about a feeling of being drained or pulled downwards. It is not a feeling of elevation.

**Hands Song:** The Hands song leans on a central note (low and middle A). The melodic motif repeats over and over from the beginning till the end. The melody is
with ascending and descending steps. The melody is very predictable with ascending and descending steps. The song has a dull and passive feeling to it.

**Crocodile Song:** The Crocodile song does not have a melody, as it is a rhythmical chant.

All five least preferred songs have repeated and predictable melodic motifs. They tend to pull the listener down to a passive state, as the musical style of these songs does not stimulate, and does not appear to elicit excited responses. This can drain and tire the listener and evoke unpleasant feelings. Such reactions have been found and will be discussed at the chapter regarding behavior.

The melodic motifs in all five preferred songs demonstrate an interesting use of intervals and notes. During some places in the songs, one can see how the melodic motif repeats, thus provide security, predictability and anticipation, and yet on the other hand, there are enough changes and surprises in the melody to keep the song from becoming redundant and meaningless. There is a balance between familiarity and novelty in the melodic lines and it kept the participants focused, tuned in and excited during these songs. In most of the five preferred songs, there is a lightness that gives an uplifting feeling. This helped the girls to sustain their activity levels as well as their positive emotional responses. It invited body movement and sustained their attention as long as they heard these songs. The songs had a flow and made musical sense. This helped the participants to be organized and feel secure in one hand, and on the other hand, they could permit themselves to feel free and let go with the music. They seemed at ease with this type of songs.

**4.5 DYNAMIC VARIABILITY**
All favorite songs have dynamic variability, with many examples within songs of crescendos, diminuendos, and both loud and soft presentation. The researcher incorporates these dynamic changes in her presentations of the songs, as far as possible maintaining consistency of presentation, in the way she sings them, and accompanies on the guitar. All least preferred songs are around mezzo piano, with some variation at times during the last part of the songs. The dynamic variability evokes energy, power, control and commitment (Amir, 1999) and all these are expressed in the most preferred songs. It changes and brings about action and freedom of expression. The limited amount of variability in dynamics in the least preferred song can contribute to the monotonous feelings of these songs.

4.6 VOCAL PLAY

All preferred songs have different types of vocal play. They may be similar to real life imitation of objects, gibberish vocalization, and changes in pitch. The Train song has sounds ‘puff puff’ and ‘toot toot’, reminding the listener of the sound of the train. The ‘toot’ sound is heard on a high octave which may provokes tension and at the same time excitement. The Spider song’s vocal play was in the change in pitch. The first time the song was sung on the typical/normal register for the song. The second time the song was sung one octave lower and more accented and the third time it was sung one octave higher than the normal register and more staccato quality was added to it. The participants usually had a wider smile during the lower singing register, but they burst into laughter or put on an even wider smile when the song was sung in the higher pitch register. There is developmental evidence (Trehub, Schellenberg, & Hill, 1997) that the octave is a natural musical feature and infants and children recognize this interval better than others do.
The Nut song has sounds resembling the sound when clicking on a nutshell ‘click, click’. The sound is produced by a special effect made by the tongue and not by actually pronouncing the sound ‘click click’. This is a new type of sound that can be pretty exciting due to its attractiveness. The Monkey song has vocal play on the syllables in the words. Some of the initial syllables in some words were dragged and some had a slight ornamentation added to them. This type of vocal play gives freshness to the repetition in the verses of the song.

The Bee song also had vocal sounds that were similar to the sound the bee makes ‘bzzz’ as well as a sound ‘oops’ to stimulate the feeling of pretending to be stung. These sounds seemed very exciting to the listeners. They anticipated each sound by widening their eyes or mouth and by intense eye gazing focusing on the researcher with extreme intensity. Most of the vocal sounds in these songs were in the higher pitches. This seemed to evoke a happy response. Their activity level rose and they seemed to want these songs over and over.

In the least preferred songs, the vocal play and sounds occurred only in two songs. The Fish and the Crocodile songs included vocal play. The Fish song had one vocal sound at the end of the song of dragging and sustaining on the first syllable of the last word in the song. Otherwise the song had no vocal plays. The Crocodile song also had one vocal sound at the end of the last word of the song, dragging and sustaining on the first syllable of that word. This was the only time there were any sounds added to the song.

The findings of this study, in particular the analysis of the music, and the emotional effects of the songs, suggest that sound effects of different vocal sounds and vocal play are important when choosing songs for girls with Rett syndrome. Songs such as
those give much meaning and fun into the music and elicit many different emotional and communicative responses in the girls.

4.7 COLORS OF PICTURE SYMBOLS

Although one participant, Aviv was choosing from orthographies (black letters on white background), the remaining six participants chose from colored picture symbols.

The colors of the pictures for the five most preferred songs were:

- Brown (Monkey and Nut)
- Black (Spider)
- Light Blue/Yellow/Black (Bee)
- Red/Yellow/Green (Train).
- Others to be included on individual basis:
  - Yellow (Chick)
  - Blue (Bird)
  - Red/Blue/Brown/Gray (Farm)
  - Red/Yellow/Brown (Butterfly).

The colors of the pictures for the five least preferred songs are:

- Gray (Rabbit, Elephant and Fish)
- Green (Crocodile)
- Pink (Hands).
- Others to be included on individual differences:
  - Yellow (Star)
  - Red/Yellow/Brown (Butterfly)
  - Green (Turtle)
Green/Brown (Tree)

It is difficult to draw any conclusions to the effect the colors may have in choice preferences. Although there are three pictures of gray color in the least preferred songs, these could be compared to the brown and black color in the most preferred song. Both the least and most preferred songs have strong colors such as green, yellow and blue as well as combination of several colors. No evidence could be found when comparing the colors of the pictures, that the participants were influenced in any way by those colors when determining choice.

4.8 SONG PERFORMANCE

Some comments can be made in reference to the style of performance the songs were sung by the researcher. Although the songs were meant as a stimuli used in the intervention for intentional choice making, once they chose, it was up to the researcher to sing their choice.

The songs were pre-composed songs with a definite form, rhythm, melody and dynamics. Even so, the songs were sung to each participant as a mother may sing to her child.

The researcher was very true to the music and its form, putting the song in the center and at the same time she was very tuned to the emotions and communicative responses the participants might have revealed in response to the song. These responses were reflected in the style in which of the presentation was performed. Meaning was incorporated into the emotional expression of the singing, almost as if a new story was told each time the song was sung. Although the tempo was kept
quite the same each time it was sung, the researcher created an illusion of altered tempi when tempo variability (ritardandos, accelerandos, fermatas and pauses) were applied into the music.

The researcher never experienced boredom singing the same songs even though the participants chose some of them endlessly. The fact that the girls responded differently each time encouraged the researcher to sing accordingly.

During songs when the girls did not respond as positively and sometimes made frown faces, the researcher attempted to ignore their response and focused mainly on the song’s musical ideas. However, on some unconscious levels the researcher was influenced by their response, which could be viewed on the videos. The above description is similar to the mother modifying her behavior to fit the expression formed by her baby. It is what Malloch (1999) refers to as ‘communicative musicality’, a narrative of combination of pulse and quality of pitch, which allows “…two persons to share a sense of passing time” (p.29).

This type of communication is vital to the mother/baby or therapist/client interaction for the emotional and cognitive growth of the child. The researcher was attuned to the girls and attempted in expressing musical ideas through the songs, in order to provoke some type of emotional or communicative responses. Musicality has its origin in vocal play and it is a most important factor in the music (Umemoto, 1997).

The text in the songs is simple and repetitive. It typically tells a simple story about an animal, food item or transportation. One cannot assume that the participants comprehended the content of the text or to the grammatical structure of the song message, but this was taken into consideration, as it was believed that the
participants were sensitive to the changes in auditory stimuli both physiologically and emotionally (Boxill, 1985; McLean, 1990).

There was a degree of variability in the way the researcher presented the songs. No tape recorder was used, as it was live presentation. Nevertheless, the value of presenting the songs live has been proven to be better than recorded music (Hooper & Lindsay, 1990). In their study they presented recorded and live songs to a developmentally disabled person. The results revealed that she responded significantly better to live music than to recorded music.

**Summary:**

Many different elements, musical and nonmusical contributed to the participants’ song preferences. Some of these elements could be found in the least preferred songs, but perhaps the combination of some of the feature was the reason for their preference. For example, a combination of fast tempi, a good melodic motif with interesting rhythmical pattern and vocal play could define the parameters of a preferred song. It is more likely that the girls perceived the whole song in a gestalt form rather than detecting each separate musical dimension (Meyer, 1956; Pavlicevic, 1997).

The five preferred songs and others down the list (except for the five, six least preferred songs) had good musical features. They were interesting and stimulated the girls. They became familiar, a place where they could sit comfortably and enjoy their power by choosing these songs. The songs were short with predictable structure. The rhythmical patterns were repeated as well as the melodic motifs. Besides simplicity
and symmetry, these songs have a strong degree of regularity, which is an important basis for stability (Pavlicevic, 1997).

In spite the repetitiveness, there were ample of changes in the rhythms, melody, dynamic, and timbre to ensure that the songs were interesting enough. The element of predictability and repetition is a strong one and is necessary for girls with Rett syndrome since there is tension that is naturally released by the songs (Wesecky, 1986). All of this organizes, and gives a form to the chaotic child.

One can detect that their breathing, hand movement and their whole body movement received a meaning while listening to the songs.

The findings also correspond with the fact that similar song preferences were found in pilot studies performed at Aalborg university and Levinsky college showing that musicality of females with Rett is similar to randomly (yet musically trained) individuals of the normal population.

Hearing a musically well balanced song can create a state in which the girls’ whole being ‘tunes’ in the music and as a result gives order and sense. The child then is available to open up to her surrounding and to engage in communicating with her environment (as will be discussed in the next section). One can see the integration of physiological organization with the psychological expression, which might have set a way for the cognition to develop and to expand. On the other hand children with Rett syndrome and other children with developmental disabilities present physical reaction that may be closer to the experience of the emotion and are not infected with rationalization (Sloboda, 1991) and in that sense their reaction can be considered a true and non-contaminated reaction to music.
It is not so surprising at all to find out that children with Rett syndrome were able to make clear preferences regarding their likes and dislikes in music, despite the evidence of significant neurological impairment, and it is safe to say that those preferences (at least in the major part) were made according to the songs’ musical elements. There is a growing body of knowledge confirming that even infants can identify categorize and group music according to different musical elements such as: tones, timbre, variations in tempo, pitch level, loudness, melodic contour, octave changes, and simple frequency ratios (Trehub, Schellenberg, & Hill, 1997).

This analysis and discussion of some of the musical elements of the songs, in an attempt to explain song preference appears to indicate preferences based on rhythmic level of musical stimulation, and tempo of presentation and is discussed in more general terms by Aldridge (1996) “…the mind and body are united within a rhythmic context of communication which enables healing to take place” (p.52).

An additional item of information to emerge from the present study (although not a research question), was that some of the girls who participated in the study used to prefer some of the non-preferred songs at an earlier age. These are songs that might be defined more as ‘relaxing’ songs, or ‘lullabies’, mostly preferred by infants and young toddlers. In contrast, most of the preferred songs can be categorized as ‘play/action songs’ highly popular with children at the kindergarten. This might suggest that at the average age of seven, girls with Rett syndrome prefer songs that are appropriate for normal children of the same age group or a bit younger. This finding is interesting and needs further investigation.

The next chapter will discuss the main results of the study (as presented in Chapter 3), and related the findings both to what is known about the population, and also the literature relating to music therapy and songs.
CHAPTER FIVE

DISCUSSION

The discussion chapter will begin with a short overview of the main findings of the study. Then, a reflection on the research design will be made, followed by a systematic discussion of the main findings of the study in the same sequential order as those findings were presented in chapter 3: Intentional choice, Response time, Learning process, Song preferences, and Emotional, Communicative and Pathological behaviors

Summary of research findings

Analysis of responses in all participants reveal that during the baseline period there was lack of intentional choice making, whereas when the intervention period was introduced (followed by periods of followed maintenance probes), confirmation of choice increased significantly in all participants.
Response times decreased, demonstrating that the participants needed longer response time during the first three to four sessions, and thereafter their response time dropped to 15 seconds or less.
The participants demonstrated learning skills within the first set of songs and between the sets, showing a reduction of trials needed to reach the pre-established criterion. Learning was maintained throughout maintenance probes and during maintenance, three months after the research had ended. Song preference was clearly established and analysis of the song structure, the songs’ musical and non-musical elements revealed that there were very specific elements to the songs that attracted the participants (see Chapter 4).
An analysis of emotional, communicative and pathological behaviors at a qualitative level showed that there was an emergence of different types of response in certain types of songs and during baseline. Some positive responses were evident, which then tailed off accordingly when the songs were repeated too frequently.

Overall, the participants were effectively choosing songs, responding appropriately to songs, demonstrating anticipation of elements and events in the songs and responding within normal response times (demonstrating learning skills), and the effects over time were significant in all cases.
5.1 DISCUSSION OF RESEARCH DESIGN

The study investigated whether children’s songs in music therapy enhanced communication skills in girls with Rett syndrome. A single case, multiple probe (multiple baseline) design was applied to answer the research questions. Multiple probe, single case design is not commonly used in music therapy research, and a discussion on this particular design and its adaptation in clinical research in music therapy, and specifically in the Rett syndrome population is relevant to evaluate it’s reliability, validity and usefulness in it’s application here.

The multiple probe design is commonly used in naturalistic environments such as classrooms with a population of people with developmental disability (Bambara et al., 1995; Hetzroni & Shalem, 1998; Hetzroni & Schanin, 1998; Hughes, Pitkin, & Lorden, 1998; Nozaki & Mochizuki, 1995; Sevcik, Romski, & Adamson, 1999). Multiple probe, single case design has also been used in studies attempting to establish communication in girls with Rett syndrome (Hetzroni & Rubin 1998; Koppenhaver et al., 2001; Sigafous et al., 1995; Sigafous, Laurie & Pennell, 1996). The multiple probe design, a time series design, is particularly appropriate when evaluating the effects of learning and its process, and this was one of the goals set forth for this study. The intermittent measurement (baseline probe) provided evidence of whether behavior change had occurred prior to the intervention. This method analyzes the interaction of the independent variable in treatment and control conditions, and enabled the researcher to identify both appropriate responses and learning over time when comparing results in the sequences of related sets of songs (Cooper, Heron, & Heward, 1987). The design is a flexible one, enabling analysis of the effects of the independent variable across multiple behaviors and/or cases without withdrawing the treatment. The typical multiple baseline design uses a very long baseline throughout the research, therefore interjecting probes during, in between and following the end of intervention sessions at equal intervals was found more appropriate for the population under investigation.

The fact that the participants in this study demonstrated impatience during baseline and baseline probe sessions, as well as “protest behaviors” (such as: crying, shouting, pushing
away picture symbols, closing eyes or turning their heads away), the use of multiple probe
design (rather than multiple baseline design), made it somewhat easier and more tolerable for
the participants. An additional cause for employing multiple probe design rather than the
multiple baseline design is that the Rett syndrome population typically has poor attention span
and their ability to take part in a session longer then 30 minutes might be limited. During the
research, the participants became exhausted when the baseline probes were applied into the
sessions. These were done with limited frequency, whereas if the multiple baseline design had
been used, these baseline probes would have been applied throughout the sessions, possibly
affecting the participants to a more serious degree in terms of their performance potential.

Previous researchers have suggested that intentional communication with individuals with Rett
syndrome should be investigated through multiple case studies. As this type of design would
allow for description and differentiation of intentional skills, group design may not be
sensitive enough to the individual differences (Woodyatt & Ozanne, 1993, 1994). The present
study found the multiple probe design to be most appropriate in collecting and enabling
analysis of data concerning Rett population’s ability to communicate and use intentional
choice making. This design has been found sensitive enough to differentiate individual
abilities and variables.

A single case, multiple probe design, was practiced in a study that attempted to teach two
individuals with Rett syndrome to request preferred objects through picture symbols (Sigafoos
et al., 1995). Prior investigations employed only one or two stimuli per trial and these were
normally the actual objects rather than picture symbols. The participants were believed to be
incapable of responding to picture symbols (Hughes et. al., 1998). In the present study two out
of the seven participants were presented with four stimuli at a time, and all were presented
with symbols rather than objects. The evidence from this research supports the hypothesis that
females with Rett syndrome can be presented and challenged with communicative demands
more than had been previously anticipated. It could be suggested that the particular attraction
of songs and music could demonstrate a higher potential for communication in the wider
population of females with Rett syndrome, if further research studies repeat replicable
findings.
It was taken for granted that the girls in this study (as they developed almost normally until the age of 18–24 months before diagnosed with Rett syndrome) did not need the actual object (which is necessary for a young developing baby) because they had previously experienced objects and pre-literacy activities (i.e. picture books read to them). They were familiar with symbols and orthographies.

Sigafoos, Laurie, & Pennell (1996) discussed the limitations of their data in terms of the lack of maintenance and generalization. In this study the first of these limitations was addressed by intermittently administering maintenance probes, after the intervention of each set was terminated. Maintenance sessions were applied at the end of the investigation as well. The maintenance sessions were performed after two weeks, four weeks and six weeks intervals, and have summed up to a total of three months after the investigation was terminated. Adding maintenance sessions to the end of the research (as suggested by Sigafoos, Laurie, & Pennell, 1996) was found most important in establishing process of learning and it’s sustainability over time (further discussed in the “learning process” section in the discussion chapter). As far as generalization is concerned, this study did not involve the use of a randomized control design, and the participants acted as their own controls. Therefore the generalization of the results has to be limited to the participants of this study, and not beyond. The question of whether the consistent improvement over time in all participants in this study, and the very large effects of treatment may support one–tailed hypotheses for future studies will be addressed when discussing the implications of the results.

Single case design is a suitable design for the therapist, as it stays close to the practice of the therapist (Aldridge, 1996). The present study, a single case design, had the flavor of therapy sessions. It took place in a natural environment, in a known setting, and in a familiar situation to the participants and the researcher. When taking into account the difficulty the Rett syndrome population has in acclimatizing to new places, people and situations, it was believed that the success they demonstrated was as a result of the familiar setting, familiar therapist and familiar materials. The familiar situation may have reduced the effects of unexplainable influencing variables, and enabled the participants to focus on the task, freeing the researcher to focus on the goals of the research.
It also has to be argued that the familiarity of this situation will have contributed the very large effect of treatment when compared with baseline, and one should not assume that the same effect to be achieved in all situations by all people. A new therapist or a change in the songs presented, or even a new setting could influence the effect of treatment. The present study had a concept of process behind it so that the participants could benefit from the actual learning experience with the intention of applying it to other situations during their daily lives.

Some ethical issues of the design should be considered in future studies. During baseline, when no songs were sung in response to the participants’ choice making, the participants on occasions seemed confused, upset, bored or angry. During the interventions, the participants’ choice was followed immediately by a song. They became frequently puzzled and confused, when baseline probes (with no music) were at times simultaneously undertaken on proceeding sets. These baseline probes may have provoked a feeling of failure as they were being sustained concurrently with the interventions, and the process of the intervention was disrupted as the probe measured periodically throughout the intervention. Sessions became very long and dissatisfying during baseline when no music was applied. They also became very long when baseline probes were applied to the intervention sessions as well as during maintenance sessions when all three sets were used. However, this was due to the fact that this researcher decided to include three sets and 18 songs (six songs in each set) in order to be certain that intentional choice making was not a coincidence in one or two sets, but in three sets with different songs. Therefore the negative effect of the baseline probes was inevitable, and it adds to the efficacy of the intervention that this did not negatively influence through carry over effects with this population. The researcher was also interested in looking at a longitudinal process of learning in children with Rett syndrome, but as a result, the sessions (when probes were incorporated) became lengthy. It is recommended that in order to avoid long baselines and sessions, fewer songs or sets may be administered in future research, providing a larger sample is used.
5.2 DISCUSSION OF RESULTS

Introduction

The participants were referred to as individual cases during the result chapter, and then some analysis was undertaken considering all the participants’ results together. While the design was made around single case examples, in the discussion the participants will be referred to as a group, in order to address aspects of the results where the entire group revealed similar responses. Nevertheless, consideration of these results will also emphasize the uniqueness and the differences within each individual, but found many similarities between the participants as a group.

5.2.1 Intentional choice making

Intentional choice making will be discussed in respect of baseline, probe, intervention and maintenance conditions. The researcher informed the participants whether or not a song would be heard after they had confirmed their choice. In spite of being informed (and also depending on whether, as individuals, they understood what they were being informed about) this strategy might have initially been very confusing to the participants. The participants had to learn a new choosing procedure. Not only was it a novel procedure of choice making, but also having to choose during baseline was not a common occurrence. The choice making procedure the participants were familiar with (prior to this study) could be perceived as a random ‘choice by chance’ (not involving confirmation), and would not have been adequate for this particular research as one of its main purposes was to investigate intentional choice making potential within the Rett syndrome population. An intentional act was considered to be intentional when the participant directed the behavior (confirming the choice in this case) towards an adult through modalities such as eye gaze, gestures, smiles or vocalization (Iacono et al., 1998). Due to this consideration, the participants had to adapt to a new procedure of choice and had to confirm the choice.
It should be pointed out that part of the method involved randomly moving the picture symbols around on the board after the participant had made a choice before the second presentation (confirmation) was made. The purpose for this was that her choice would not become habituated, but rather derived from understanding the concept of intentional choice making. This random presentation of symbols was decided partially due to a comment made by Olsson & Rett (1985), warning against over estimation of functioning level in the population of Rett syndrome due to repeated experience. The results demonstrated that girls with Rett syndrome have intentional choice making abilities. As the girls’ intentional act had been confirmed and established during this research, the findings suggest that it would be adequate to request a choice only one time and accept it as an intentional choice.

**Baseline - all participants:**

An inconsistent and very low rate of intentional choice making was revealed during baseline. There are two possible explanations for the low rate of intentional choice and the inconsistency in choice making. A non-responsive reaction by the participants may have been the result of their possible comprehension that they would not be receiving a song despite their choice making (this was announced prior to their choosing). This comprehension and the response accordingly are referred to by McLean (1990) and correspond with the intentional communication stage.

Low rates of intentional choice making might also be explained by the fact that the procedure of choice making was new and unfamiliar to the participants. Normally, they experienced making a one-time selection and immediately thereafter being rewarded. It should be noted however, that the improvement in choice making ability observed during the intervention over time and through experience, did not occur at baseline, where a negative inclination and inconsistency was noted. This phenomenon weakens the possibility of novelty of this type of choice making procedure as the cause for diminished choice making ability. It rather puts the responsibility of low choice making rates on the fact that the participants did not receive a song after they had chosen it, they were frustrated, and showed low levels of tolerance.
The low level of intentional choice making was a common factor between all the participants. This finding raises some thoughts regarding the level of understanding of people with Rett syndrome, which demands further investigation.

All participants showed some type of negative response during baseline. This will be discussed in the emotional/communicative/pathological behavior section.

**Baseline Probes - all participants:**

The baseline probes in sets 2 and 3 showed low levels of confirmation of choice, thus very low levels of intentional choice making were demonstrated during the baseline probes. The same reasons as in the baseline (not receiving a song for their choice making), may have led to the low rates of intentional choice making observed in the baseline probe sections in all participants. At times some of the participants showed more confirmation than anticipated. It could be due to the fact that the probes were carried out on the same sessions and in the same situations as the intervention, which may have resulted in confusion. Other participants (maybe higher functioning) revealed significantly less confirmation during the baseline probes as they responded less during baseline probes.

**Intervention - all participants:**

The difference between the baseline, the probe and the intervention sections were evident. Once the intervention was introduced (the songs were added to the study), a drastic change occurred. Introducing the songs enhanced the participants’ intentional choice making and brought it to its potential. This was clearly displayed by their ability to confirm their choice, making it intentional. Choosing was consistent for all choices presented both for when first asked to choose and when repeatedly asked to confirm their choice. Their ability to make intentional choices was evident and was almost fully expressed at that point.
All girls made a drastic change during intervention, however it typically took a gradual form starting with average intentional choice making at the beginning of intervention (choice making between 1-3 trials of the first set). It then improved steadily and constantly up to a very high level of intentional choice making by the third session of set 1 and onward. After the participants had reached the criterion of set 1 and a new set was introduced, (set 2 and thereafter set 3), their intentional choice making dropped during the first session in every set. Typically, during session 1 sets 2 and 3 confirmation fell behind, but then became stable during session 2 of each set. This could be explained by the novelty of the new set of songs being presented. A change occurred and a new set was introduced with six new songs (some familiar and some un-familiar songs). The transition from one set to the other required a period of familiarization. The graphs during the intervention period in all participants reveal a consistent learning curve. This fact will be further discussed in the section concerning learning ability.

The fact that during baseline the participants showed negative responses, while during intervention they exhibited very positive responses, will be discussed in the emotional/communicative behavior section.

**Maintenance - all participants:**

All participants achieved consistency in intentional choice making during maintenance sessions at a very high rate (97.5%) steadily increasing from set to set. It was clear that intentional choice making was enhanced by the introduction of the songs (intervention and maintenance). The maintenance sessions showed almost no individual differences as the participants (as a group) chose intentionally at very prominent levels.
Summary:

The participants in this study, girls with Rett syndrome, revealed a strong ability to choose songs, then confirming their choice, demonstrating intentional choice making. Most girls had been previously exposed to the selection of songs during group music therapy. Four girls, Aviv, Hilla, Meirav and Talia, had a longer experience over four years (once a week) during 40 minutes group music therapy sessions with the music therapist who was also the investigator in this research. The remaining girls, Elisheva, Rachel and Tali had less experience, amounting to about one session a week for one year of 30 minutes group music therapy with another music therapist. All girls may have received one opportunity during the weekly music therapy session, and each week her choice repertoire may have been different from the previous weeks. It should be questioned if such exposure can be considered an experience in choice making. All methods of choosing during these sessions were different from the one that was employed in the study. In these weekly group therapy sessions, they were requested to choose only once the song of their choice, and then the song was immediately sung to them. The procedure in this research demanded confirming the initial choice in order to verify the intent. As choice making was not necessarily a primary function or learning requirement in therapy, the consistency was correspondingly erratic. However, it has to be recorded that the participants had previously been exposed to, and were engaged in activities where songs were offered for choosing, and may have had a positive influence on the result, limiting the conclusions one can draw from the large effect of treatment.

These findings clearly show that the participants in this study have intentional choice making ability, and further investigation is warranted to determine whether these very positive results would generalize to the wider population. It is also apparent from the findings that these abilities are improving over time. Due to repetition and practice, it is evident that those abilities (after being achieved) can be sustained over periods of time (lasting up to three months) despite the fact that the intervention was withdrawn for periods of time during those months. This suggests that music, and songs in particular, have an important role in revealing such potential in a population that until not long ago was thought of as uneducable, and with pre-intentional communication.
Considering the way this study was undertaken, these results also suggest that an attentive cooperative child, a good rapport between child and researcher, a familiar situation, and strong motivational factors will facilitate such positive outcomes.

5.2.2 Response Time

Measuring response time was undertaken during the trials, as an additional source of data not directly related to the primary hypothesis. Evaluation of the typical response time in other studies tends to report that girls with Rett syndrome have delayed, sometimes severely delayed responses. The difference that was observed between the present study and previous ones, namely the changes in response time observed within the current research, have made it an area that merits closer analysis.

Most other studies report a 30 seconds time interval to allow response in children with Rett syndrome as they often require this amount of time to initiate a response (Lindberg, 1991; Sigafoos et al., 1995, 1996, 1999). Koppenhaver et al. (2001) suggested a shorter response time interval of 10-30 seconds. The current study established a criterion of a 15 second interval of response time as an appropriate time band for each opportunity of choice making, as the researcher felt that once the participants became accustomed to the study, the allotted time would be adequate.

The response time was timed during choice making opportunities throughout the study. The intervals were set up in a five-step choice making opportunities for each time the participant was asked to choose, and a five-step opportunity when confirming their choice. The participants were given 15 seconds to respond for every opportunity before moving onto the next step. Only the first two steps (less than 30 seconds) were considered and counted into the study, as the remaining three steps all had some degree of cueing and/or prompting (see procedural section in Chapter 2). The rational behind enabling the participants to have five opportunities was to allow for many learning opportunity experiences.
Even if making a choice was a difficult task for the participants and after five steps they would not have made one, they eventually would have had the opportunity to hear the songs.

**Response Time - all participants:**

The response time during intervention period was selected for analysis. Intervention had taken place after 4-5 baseline sessions and by that time the participants had become familiar with the choosing procedure. Their motivation to choose was at its highest when they received the song of their choice.

Results show that all of the participants in this study initially had a need for at least 1-3 bands/steps (between 15-45 seconds) and some, even more. From the third or fourth session they all required only 15 seconds or less to respond to the stimulus.

As found in previous studies, all participants demonstrated delayed reaction during the initial trials. This was consistent with the findings of Woodyatt & Ozanne (1994) where the girls’ response time was often delayed and commands needed to be repeated. In the present study it was found that time and experience was necessary to lower their response time.

The participants seemed initially unsure of what was expected of them, however over time their performance became faster. Sigafous et al. (1996) reported that girls with Rett syndrome’s response to choices of picture symbols became functional during intervention, while exploratory during the baseline. Their report seemed to correspond with this study from the viewpoint that the participants were functioning on target when responding to the stimulus during intervention and during baseline they were unresponsive to the point of rejecting the stimulus.

They responded with ease and happiness to the choice making activity during intervention. It should be noted that the response time became somewhat slower during maintenance. This could be due to the time lag between the end of the intervention period and the beginning of the maintenance. This could strengthen the notion that it is vital for this population to have many opportunities for different response activities throughout the day involving choice making on the basis that the more they do it, the more proficient and immediate is their response.
The findings in this research revealed that children with Rett syndrome initially demonstrate a delayed response time when the intervention is short term, or when considering only the scores at the beginning of an intervention when the girls are still learning what they wish to choose. When there is constantly practice and rehearsal, the results of response time will improve. These findings are congruent with the description of the Rett syndrome population so vividly presented by Lindberg (1991), and with the fact that children with Rett syndrome suffer from apraxia (a dysfunction in motor learning and motor execution), a condition that can be improved by constant repetition, until the action becomes automatic.

The reduction in response time is due to proper and meaningful interest (and as a result a proper response) that has been achieved when a strong motivational factor, such as music (songs) was used. The stimulus in this research (songs) was just such a motivation factor and the girls seemed eager to respond to the stimulus.

A second factor to have influenced the decrease in response time was probably the time that elapsed from the beginning of the research and allowed the participants time to learn the new procedures (this will further be discussed in the section regarding learning process).

If the assumption presented above is correct (that children with Rett syndrome need more time to learn new procedures before achieving their potential), then this might mean that with every intervention employed with this population, a preliminary intensive acclimatization period is needed (in this study three times per week over a 2-3 weeks period) before the achievement of a basic skill level for learning. This needs further investigation.

The point that a child reacts differently (better) to the person and situation they knew in an examination situation (as was the case in the research situation) should be taken into account. Recommendation arising from this study that should be considered in future studies with the population of Rett syndrome is the need for an introduction period, enabling the child to familiarize with the situation, setting and the person involved in the intervention.

In a recent reunion with five out of the seven participants (two years after termination of this study), they were given opportunities to choose songs. Their response time was initially around 30 seconds and within the second and third opportunity, they all responded less than 15
seconds. It seems that they had all maintained the learning of this activity and as a result responded eagerly and within a very short time period.

**Summary:**

The study was set up in a five bands/steps hierarchy, allotting 15 seconds for each step. During the research these were measured in units of 15 seconds. The participants initially needed between 15-45 seconds before they made their choice (up to 3 steps), in other words; there was delayed reaction, which was demonstrated by nearly all of the participants within the first 3-4 sessions during the intervention. Immediately thereafter, the participant’s response time diminished drastically, and they all chose within the first 15 seconds (step one). Although previous research showed delayed reaction (Woodyatt & Ozanne, 1994), this study initially had similar results, but within a few sessions the participants demonstrated no delayed response. The contrary occurred, as several times the participants indicated their choice before the researcher had managed to ask them to do so.

The researcher assumed that some of the participants would have needed many trials, and therefore the five-step hierarchy system was set forth. Fortunately, the research protocol was established in a way that the participants did not have to go through all 5 steps.
5.2.3 Learning process

The learning process was analyzed within and between sets of songs and during maintenance sessions.

The present study was set up in a way that learning could take place. The duration period needed to complete the research was unknown (as described in the methodology chapter). Furthermore, it was assumed that some participants could end earlier than others. The study took into consideration individual differences because of natural circumstances and learning abilities. The results demonstrated that all participants completed the study during the same week within one or two sessions apart. It took every participant about the same amount of time to reach the criterion of each set. Learning process took place and was analyzed within the first set of songs, between the sets and during maintenance. The participants were not familiar with the type of choosing procedure, and naturally this novel procedure demanded acclimation time.

**Baseline:**

No learning had occurred during baseline. This was due to the fact that music was not introduced during baseline hence, no motivation was set forth to enhance learning.

**Intervention:**

All participants began learning and demonstrating a steady ascending trend of choice making during the first set, at the point of being exposed to the music. Aviv, Meirav, Hilla and Talia had greater past experience in choosing consequently, it was postulated that they may have been confused, having to make the change from the familiar and known choosing procedure, to a new more complicated one. Aviv had difficulty during set 1 in confirming her choice, and as a result needed six sessions to complete that set. Her steady learning can be noted, and as she reached session four she began to comprehend what was expected of her. The same process was observed in Meirav and Hilla who needed five sessions to complete set 1. Talia needed seven sessions to complete her first set. It was believed that all four participants
needed this amount of time as a result of change in choosing procedure. This phenomenon, observed in the experienced learners (during the beginning of intervention) should be taken into consideration by future researchers in the field of cognitively impaired individuals in general and the Rett syndrome population in particular.

Elisheva, Tali and Rachel were less experienced in choice activities as the former four girls. Their adjustment to the method of choice may have not been as confusing as for the others. Even so, Elisheva needed six sessions to complete set 1. Rachel and Tali reached criterion after only four sessions. They had learned very quickly the procedure of choosing. Elisheva had extremely low abilities in choice making during the initial two sessions. It was very difficult for the researcher to understand her choice, to the point that the researcher had to lay on the floor in order to see what symbol she was focusing on. During the third and fourth session her choices became much clearer. This example illustrates the value for the researcher to be well informed and experienced with a clinical population. The present researcher had also worked as the music therapist in the day care facility attended by the children and her knowledge of Elisheva helped in understanding her reaction. It has been known that children with Rett syndrome are slow adapters, and Elisheva took this trend to the extreme. Whenever Elisheva went out of her classroom to the playground she normally cried for 5-10 minutes and the same occurred on return to the classroom from the playground. Staff interpreted this behavior as representing Elisheva’s distress at the transition from one situation to another, a characteristic that can be identified in common with childhood autism, with which girls with Rett syndrome were formerly assumed to have pathological characteristics in common. Without personal knowledge about the participants, a risk of different analysis of raw data may exist.

All participants except for Rachel and Tali reduced the amount of sessions in set 2 after all the practice they had made in set 1. All participants but these two fulfilled the criterion in set 2 faster than in set 1. It seems that learning can be demonstrated as a result of this achievement. Two factors may have contributed to Rachel and Tali’s increase in sessions during set 2. One factor may be due to the fact that they both had been away from school due to an illness during set 2. This disrupted the flow of set 2. The second factor could be that during set 1 they both reached the criterion very quickly, not enabling enough time to stabilize choosing during set 1,
whereas the other participants had two more sessions (more choosing opportunities) than they did. Both factors should be taken into consideration. Such events can teach us two things: The first emphasizes a point presented earlier, that a researcher who is also a clinician and is familiar with the subjects can be aware of influencing factors such as illness of the child that may influence the flow of the intervention. The second point is that taking pauses from any activity or practice, especially choice making activities may disrupt the flow for advancing in girls with Rett syndrome’s abilities, as is the case in choosing. Those incidents suggest that many consecutive consistent experiences of choice making are needed in order to internalize learning in this population. Such findings are in agreement with previous reports stating that learning choosing skills and establishing preferences is of utmost importance for people with severe disabilities. “If expressing preferences and making choices are expected to serve a function in people’s lives and extend their self-determination, quality of life and community participation opportunities to exercise these skills must be incorporated into the environments in which people live, work, recreate and attend school and throughout their day-to day activities” (Hughes et al., 1998, p.313).

The transition for all participants from one set to the another seemed to cause some difficulties, expressed in a drop in their intentional choice making during the first session in the new set. This can be due to the novelty of the set and of the songs presented, and as a result more time was needed to determine a choice. During the following sessions, they all chose with very high intentional levels.

**Maintenance:**

All participants demonstrated learning during maintenance. The long gap that occurred between the end of the intervention till the beginning of the maintenance sessions and between each maintenance session should be noted. The least amount of time was two weeks, then four weeks and six weeks between each maintenance sessions. This almost contradicts the previous statement that taking breaks may disrupt the flow, since the results reveal that the opposite occurred during maintenance. This could tie into the second statement that many experiences are needed for learning to be internalized.
By the time maintenance sessions had begun, the participants had a vast amount of experience in choosing throughout three sets of songs, as well as during the maintenance probes, sets 2 and 3. It seemed that the participants must have internalized what they had learned as they kept their choosing scores at such a high level. The notion that the girls internalize what they have learned became even more prominent as the researcher recently met five of the participants and gave them opportunities to choose same songs as in the study. They all chose at a very high level two years after the study had ended.

**Summary:**

Each participant began the study from a different point of experience. Some had more experience than others did. This may not have always made a difference during the study. The participants demonstrated that they all had the ability to learn and made an impressive progress within and between the sets. By the time maintenance sessions had begun, intentional choices between songs were performed at the same level in all participants.

The capacity to learn new skills when provided with opportunities has been postulated by a few professionals and scientists in the field of Rett syndrome (Jacobsen et. al., 2001; Kerr, 1992; Leonard et al., 2001). There are several types of learning and some authors have postulated that the population with Rett syndrome might have the ability to learn through "basic forms of learning" such as conditioning (Hogg & Seba, 1986; Remington, 1996; Rescorla, 1988, in Demeter, 2000). In the present study learning took place despite the fact that the participants had to learn a new form of choosing procedure and despite a constant change in the position of the symbols - and not by conditioning learning.

An important basis in the learning process was the fact that the girls established a relationship with the therapist as few clinicians/researchers have suggested before (Hill, 1997; Montague, 1988; Wigram, 1991; Wigram & Cass, 1996). The researcher had previously worked with four of the girls and was a familiar figure to the remaining three. The participants were comfortable with the researcher, her style of interacting and singing, and as a result were available to participate in a new experience and at the same time learn.
The success in learning through songs in music therapy may have been a strong motivating force, made a change and was a strong ground for learning (Boxill, 1985). The girls were familiar with most of the songs and were eager to hear them sung by the researcher.

### 5.2.4 Song Preference

The research hypothesized that girls with Rett syndrome can make choices and are able to express song preferences. It could be drawn from the results that girls with Rett syndrome can choose and have a clear preference for certain songs. The participants demonstrated choice by selecting a song symbol, consequently making an act to receive a song. Their preference was inferred from the act of choosing.

The percentage of choosing a song was calculated for each participant. The higher the number of choices made, the higher was the preference level of that song. The lower the number of choices made, the lower was the preference level of that song. In this study, eighteen songs were presented during the investigation, in which twelve were familiar and six were unfamiliar songs. They were divided arbitrarily into three sets of six songs each, four familiar and two unfamiliar (more detailed in the material section in Chapter 2). The results reveal that the participants have individual song preferences. The five most and the five least preferred songs will be discussed in more details.

The five most preferred songs of the group were the Nut, Monkey, Spider, Bee, and Train songs. The least preferred songs were the Rabbit, Elephant, Fish, Crocodile and Hands songs (musical scores of all songs are presented in Appendix XIII). This was determined by calculating the choice making percentage of all songs.

Three of the participants had their own individual song preference. Aviv preferred the Train song, Hilla the Monkey song and Meirav the Spider song. Elisheva and Talia shared the same preferred song, the Bee song, and Rachel and Tali shared the same preferred song, the Nut song. Although each had their own preference, all of these preferred songs were the top five preferred songs.
Despite the same tendency of preference for all participants, it is clear that each one had her individual preferences. It is interesting to note that during the first four favorite songs some participants chose similarly, but on the fifth preference each participant had her own individual song preference.

All of the above favorite songs were chosen to a very high percentage. It was clear that girls with Rett syndrome in this study have strong preference for certain songs, and the same was true for the five least preferred songs. Aviv, Elisheva and Meirav’s least preferred song was the Rabbit song. Hilla and Talia’s least preferred song was the Fish song. Rachel and Tali’s least favorite song was the Elephant song. All of these songs were grouped into the five least preferred songs. During the first three least preferred songs there was more of a consensus in the group, but during the fourth and fifth least preferred songs each participant had their own individual different song.

There was a very big difference in the amount of times each participant chose her most preferred songs in comparison to the amount of times she chose her least preferred song. Also, their emotional and communicative behaviors were very different when the most and when the least songs were sung to them. These will be discussed in the next section.

The findings referred to above, demonstrate the fact that females with Rett syndrome as a group have discrete musical preferences (in this case song preference) and each child had clear individual likes and dislikes. This information coincides with other reports (Holdsworth, 1999; Merker et al., 2001; Wigram, 1991, 1996; Woodyatt & Ozanne, 1994), although the present study was able to show those preferences through scientific observation and measurement. The importance of such perceptions in the fact that establishing each child’s individual preference is an important first step when trying to build a proper motivational factor list to ensure cooperation of the child in any activity calling for active participation.

The findings in this study can contribute not only to the Rett syndrome population but also to a wider range of developmentally disabled children. Hughes et al. (1998) have found through
their review of 27 studies assessing preference and choice in people with severe and profound disabilities that they have: preference, it can change over time, and may vary among individuals.

Nozaki & Mochizuki (1995) reported that a woman with severe disabilities preferred children songs and tunes rather than jazz music. It is therefore vital to assess the variability of preferences, as these can change and be affected due to environmental contingencies and individual reinforcement history.

Four of the participants in this study (Aviv, Meirav, Hilla and Talia) who in the past responded extremely positively to the Fish and the Elephant songs indicated no pleasure when hearing these two songs. These were their least preferred songs during the present study. The girls were at the time when they liked the songs much younger and these songs may have suited them then at a younger age (toddlers). These songs are quite slow and may remind the listener of a ‘lullaby’ song. For these girls the change in preference may have been due to the girls' development into more sophisticated children's songs and age appropriate.

The value of indicating preference, to accept or reject an item is of much value for this population as it can be “…the path to improve quality of life and to self-advocacy” (Nozaki & Mochizuki, 1995, p.201).

As an informal ‘follow-up’ to this study which had revealed such clear agreement between the participants for preferred and non preferred songs, two small experiments with non-clinical populations were carried out with two different groups. One group was composed of ten music therapy students from Levinsky College in Israel, in their final year (where the researcher had been teaching). The second group was ten Ph.D. music therapy researchers and professors at Aalborg University in Aalborg, Denmark where the researcher studies. All ten songs were performed (by the researcher), just as it was during the study. The order in which the songs were sung was arbitrary. The participants were asked to rate all ten songs from most to least preferred songs. Both groups rated the songs in exactly the same order of preference and non-preference as the clinical participants in the original study (the group with Rett syndrome).

The results of these small studies may imply that the participants in the original study had the ability to differentiate between the songs.
This strengthens and validates the point that the group of girls in this study has a quite individual and definable taste in music when compared with a group of non-clinical subjects. Those finding co-inside with previous findings by Trehub (1985, 1987; Trehub, Trainor, & Unyk, 1993), which reported that small babies and infants have individual musical preferences and differences and that those relay on the ability of such populations to differentiate musical elements in an adult like way. In addition, it is worth mentioning that the second group in the pilot study (the Aalborg University group) consisted of people from various places/cultures in the world and yet their preference list was similar to the group of girls with Rett syndrome. The same was true for the first group in the pilot study (Israeli students from Levinsky College). Most songs were American and British children’s songs translated into Hebrew while the researcher composed the remainder songs. The songs had been used for many years prior to the study by the researcher and had been very popular among American children (both normal, and those with disabilities) who came from many different cultural backgrounds such as: Puerto-Rican, Chinese, Irish, Middle Eastern, African-Americans, Native-Americans and Canadians. Taking all those facts into account, including the fact that the study took place with Israeli participants and the first pilot study was carried out also with only Israeli students, may suggest that the songs have a ‘universal’ quality in them, as yet undetermined in the analysis.
5.2.5 Emotional, Communicative and Pathological Behaviors

Emotional and communicative behaviors in the present research were analyzed according to previous reports made by Sigafoos et al. (1999, 2000) on research projects with the Rett syndrome population. The present study will expand and enhance these non-formal ‘Potential Communication Acts’ with and attempt to attach some meaning to these behaviors. In the present study each girl exhibited different behaviors, unique to herself while many similar emotional and communicative behaviors can be viewed in the girls as a group.

The following inventory: ‘Identifying Potential Communicative Acts in Children Developmental and Physical Disabilities’ (IPCA) was described in Sigafoos et al. (1999).

- Stereotyped hand movements (wringing, rubbing, or clasping of hands)
- Eye gaze (looking at staff person for at least 3 seconds)
- Hyperventilation (rapid audible breathing)
- Vocalization (any vocalization other than breathing)
- Facial expression (smiling or frowning)
- Body movement (wiggle, kick, moving head or torso forwards or back).

Sigafoos et al. (1999) noted that the girls in their study exhibited high levels of eye-gaze, hyperventilation and stereotyped hand movements. These behaviors occurred during interaction in ‘probes’. Although the girls in their study showed high levels of stereotyped hand movement and eye gaze during social interaction, there were less of them during ‘protest probes’. Their findings are similar to the ones in this study where most girls avoided eye contact and reduced hand movement activities during baseline and baseline probes (no music). These were intensified during intervention and maintenance sessions. The eye contact during the musical interaction coincides with Wigram (1991) where he reported that girls maintain very good eye contact during music therapy sessions.

Only one girl in the present study Tali, demonstrated decreased hand movements during music. This may be due to the fact that she was rocking and head swaying most of the time during music, maybe as substitute to the hand movement. Once music was stopped, her
rocking and head swaying stopped, and an increase of stereotyped hand movement was evident.

Woodyatt & Murdoch (1996) discussed the fact that when presenting girls with Rett syndrome with auditory stimuli it provoked changes in their breathing patterns, disrupting their cycle of breathing and causing apnea periods and changes to the depth of the breathing. Hilla’s and Elisheva’s breathing cycle was changed when given the opportunity to choose and during music. They were the only two with abnormal breathing patterns. They both held their breath while choosing (in between songs), and during least preferred songs. Their breathing became shallower during preferred songs. The change in breathing patterns may be due to their interest or disinterest in the activity. The more excited they became, the less breath holding, the more hand movement, the less excited the more breath holding and less stereotyped hand movement. This postulation should be considered advisedly, when taking into account the differences that can be found between girls.

Body movement (forwards) occurred in several participants during the present study. Aviv and Rachel leaned forward at the end of a song. A few times, Aviv leaned forward and tapped on the researcher’s leg when the researcher stopped singing and began coughing during the song as if requesting for the singing to go on. Several girls pushed the picture symbols away during baseline, indicating some sort of protest. So there is some evidence that for some participants, the way they consistently (consciously or unconsciously) moved their body to indicate a communicative act or response.

The behavioral acts in this study were divided into three categories (as described in the result section of this study):

- Emotional behaviors: smile, laugh, serious face/frown, cry, moan and vocalization
- Communicative behaviors: eye contact; look away, close eyes, eye shift (avoidance), body movement (rocking of torso or head), leg movement, leave seat, walk towards door, lean forward, lean back, push picture symbol away, lean hand on researcher’s leg, put legs on researcher’s leg, open/closure of mouth.
Pathological behaviors: stereotyped hand movements, teeth grinding, hyperventilation and change in facial color.

This division of categorization was made in order to organize the chapter and to ease the reading. For this discussion however, all acts or behaviors will be gathered into one category strengthening the concept presented earlier by Sigafoos et al. (1999, 2000) that all acts/behaviors/emotional responses, made by a non-verbal child can be interpreted or referred to as communicational.

**Behaviors during baseline:**

Different behaviors demonstrated by the girls during baseline were as follows: Serious face/frown, cry, no smile and shout, turning the head away, eye shift (quick eye movements from side to side translated by the researcher as avoidance), leave seat, walk towards the exit door, push picture symbols away, look away (no eye contact), close eyes, move body backwards, stereotyped hand movement, abnormal breathing patterns such as breath holding and teeth grinding.

**Facial expression:**

One could interpret some similarities in responses from the facial expression and vocal expression of the participants. Aviv and Meirav began crying and Meirav shouted much of the time, therefore their facial expressions reflected either distress or irritation. These behaviors seemed to enhance with time, became more frequent and stronger. Aviv and Meirav have in general a wider palette of observable emotional expressions. No wonder these were expressed during baseline (and later during intervention). None of the participants revealed any positive emotional responses during baseline in their facial expression. Although the baseline was still a situation of quite intensive social interaction (a one to one situation, which the girls normally enjoy), the girls seemed to react ‘negatively’ when they received no songs in response to making a choice.
Eye and head movements:

All participants either looked away, turned head away, closed their eyes or shifted eye contact. Aviv, Elisheva and Hilla kept abrupt eye contact from time to time, but these were accompanied by a frown, serious face or cry and lasted for a few seconds only. It was interesting to notice that these three girls hardly closed their eyes, but instead looked at the researcher straight into her eyes as if confronting her by indicating their unhappiness. Hilla did not maintain eye contact for very long with, but had a very stern look/gaze in space and moved her body backwards. These responses suggest that the girls did not accept a situation where they received no song, and reacted in a way that can be perceived by a trained clinician/therapist as a communicative act. The researcher assumed the girls might be saying something like “I do not understand why I have to show you the same symbols again and again when you don’t sing me any of the songs. It is not acceptable and I would like it to stop” (intuitive comment)!

The above discussion contradicts findings that the girls accept and do not protest when they receive an item that they did not choose (Sigafoos, et al. 1995).

The present finding corresponds with a previous study (Iacono et al., 1998), stating that: “Individual signals, which occur frequently and with interpretable functions, provide some clarity to communication, despite a lack of conventional signaling” (p.111).

While closing the eyes and looking away are behaviors that might be interpreted as somewhat negative and their meaning can be debated by different viewers, there seems to be no doubt that getting up from the chair and standing at the exit door or pushing the symbols away are obviously signs of revolt and rejection. All of this when combined with previous findings display a rather clear image of dislikes and discontent changing an emotional/behavioral acts in to communicative statements.
Behaviors typical of Rett syndrome (pathological behaviors):

Behaviors typical of Rett syndrome such as stereotyped hand movement, abnormal breathing patterns such as breath holding and teeth grinding might also be interpreted as communicative acts. The stereotyped hand movements were different in each individual case. Aviv, Elisheva, Hilla, Rachel and Talia’s hand movements were present on a lower level at baseline than during intervention. These typically were smaller, slower and less amount of strength went into the hand movements. The hands were almost passive, occurred on the lower part of the abdominal, and were interpreted by the researcher as if indicating boredom or loss of hope. Tali’s hands became clenched while Meirav’s became stronger, placing her hands forcefully inside her mouth. Therefore Tali and Meirav’s hand movement didn’t necessarily become bigger or stronger; their form of expression had changed.

Woodyatt and Ozanne (1994) described that in one of their subjects hand movements increased in speed when she was agitated. This behavior is presented in other literature on Rett syndrome (Lindberg, 1991; Hunter 1999) suggesting that in this population stereotypical hand movement may represent inner feelings. Thus, the finding in the present study, that shows that most participants presented low level of hand movement is not surprising and corresponds well with cases described by other researchers and clinicians in the field.

Elisheva and Hilla demonstrated apnea. These were quite strong during the baseline. Talia was grinding her teeth, and these were very strong and loud during baseline.

Summary:

Facial expression such as: frowning, lack of smiling, lack of eye contact, lack of vocalization, and the appearance of body movements such as walking towards the exit door, or leaning backwards, were evident in all participants, and were judged to be congruent with the feelings the participants exhibited during the baseline. The hand movements seemed to have a definite role during the baseline and the intervention (as will be discussed further on).
During baseline, hand movements were less prominent in most of the girls, possibly indicating a less stimulated mood. Crying, breath holding and teeth grinding were not as common phenomena among the sample population for this study.

**Behaviors during intervention - least preferred songs:**

The behaviors expressed and analyzed during intervention in the least preferred songs were: smile (on & off), serious face, moan, eye contact, body movement (rocking, head), leave seat (go to exit door), make faces (squinting eye-brows and lips) stereotyped hand movements and teeth grinding.

**Facial expression:**

All girls smiled (on and off) as well as possessing a serious face. Three out of the five least preferred songs were unfamiliar to the participants and this type of emotional response (on/off smiles) may indicate uncertainty while hearing the songs or a mixture of emotions created by the fact that a song the girl chose was being sung to her, while disappointment from the actual song itself as a musical entity. The serious facial response may have expressed intensity and concentration when listening to the songs, while the smiling face may have indicated parts of the songs they might have liked. The serious face could also indicate dislike towards the song, but not serious enough to begin crying or shouting, although Talia was moaning at times.

**Communicative behaviors observed during the least preferred songs:**

All girls maintained eye contact during least preferred songs, which was the most frequently noted change that occurred when comparing between no music (baseline) and hearing a selected song, Tali continued rocking and moving her body, while Elisheva rocked from time to time. Aviv got up several times and went towards the exit, only to return at the end of the song when told she could choose another song.
There seemed to be much less body movement during the least preferred songs. This could be due to the fact that the girls were not engaged in really listening to the songs, as several of them were unfamiliar. Aviv was the only one who protested against these songs through her body language. Also, the minimal body movement during the least preferred songs could indicate that the girls were actually listening intently to the songs. While familiarity with the nonverbal, body language of these girls allowed some degree of interpretation where there was some consistency in response, over interpretation of body language is something to be avoided in this study, given the more concrete evidence of preference.

**Behaviors typical of Rett syndrome (pathological behaviors):**

All participants’ level of stereotyped hand movements was lower during the least preferred songs than during the most preferred songs, and a few girls stopped hand movement completely at times. Their arousal level was lower and this may suggest the reason why the frequency and intensity of hand movements was reduced. During the least preferred songs, Talia grinded her teeth. This could indicate her disliking towards the songs, as this was never the case during the most preferred songs.

**Summary:**

The response shown by all girls as a group, when listening to non-preferred songs was evidently different from the response shown during listening to preferred songs. The behaviors and emotional reactions are ambiguous, limited and almost non-existent when non-preferred songs were sung. When looking at the tables of those elements in the research, what mostly catches the eye, is the lack of reactions caused by the duality of emotions perceived by the child.
Behaviors during intervention - most preferred songs:

The behaviors observed and analyzed during the preferred songs were: smile, wide smile, laughter, vocalization, eye contact, body movement (such as rock, lean forwards), leg movement, lean forwards (at the end of song), tap on the guitar (at the end of songs), look away (at the end of songs), leave the seat, move around and return to seat (dancing?), lean hands on researcher’s legs, put legs on researcher’s legs, mouth open/close and change in facial color and stereotyped hand movements.

The most prominent fact obvious to the researcher was the abundance of responses displayed by the participants when preferred songs were played.

Facial expression:

All girls demonstrated these responses. Tali had never been observed laughing outside the research setting. She typically smiled widely but seldom laughed. Talia, on the other hand, had shown laughter in many occasions in the past. Talia’s mood had not been very positive for the past several months (prior to the study). This could be due to her deteriorating physical condition during that period. It is quite common for this population to indicate and stress their emotional and mood states according to their physical status (Lindberg, 1991). It is therefore advisable to know their physical condition when working with the girls. Not withstanding possible discomfort due to a physical cause, Talia demonstrated happiness by smiling widely and vocalizing during preferred songs.

Aviv, Meirav, Tali and Rachel were quite outgoing, at times quite loud during preferred songs. Although Tali did not laugh, she shouted and vocalized (singing?) while smiling. Hilla and Elisheva were more reserved, although demonstrated all of the same behaviors. All participants seemed happy and appeared to displace more freedom in expressing emotions when hearing preferred songs.

The girls emotional response coincides with Latchford (in Trevarthen & Burford, 1995) who implied that even severely impaired children react appropriately by smiling and laughing when
being joked with or teased. Despite the disability they are left with some level of intact emotions at a subtle level.

**Vocalization:**

All girls vocalized somewhat during the preferred songs, but nearly not as much as the researcher had anticipated. This hypothesis came about as a result of the amount of the girls’ vocalization during improvised individual music therapy. There was much more space and freedom in the music during improvised music therapy and they vocalized more during these sessions. It seemed that the girls did not have adequate time to produce vocalized response during the structured songs in the research. They were more active listeners. The results of relatively low vocalization in the present study are very similar to the findings of Sigafoos et al. (1999). In their study during structured probe activities their participants hardly vocalized at all.

In the present study, the participants vocalized more during most preferred songs than during less preferred songs, but these were relatively low. An interesting phenomenon occurred. Vocalization increased in quantity and intensity after the song had been sung, and mostly at the end of the sessions. Stern et al. (1975) discuss two modes of communication: ‘co-action’ (mother and infant vocalize together) and ‘alteration’ (mother and infant alternate vocalization). The mode of alteration was how the girls in this study responded. They concentrated on the therapist while the song was sung to them, as a young child may be when listening to his mother talking ‘motherese’ or singing to him. The girls in the study moved their bodies to the singing as the baby will move in synchrony to the mother’s voice (Burford & Trevarthen, 1997; Stern, 2000; Trevarthen, 1996; Trevarthen & Burford, 2000).

Clair (1996) reported similar findings in a study with the population of senile dementia. In that study the participants kept quiet whenever the researcher was talking or singing, and initiated verbalization only when there was silence. It could have been that during the present study the girls translated the silence parts as their turn to participate in the conversation and hence began to vocalize.
The findings suggest that in order to elicit or enhance girls with Rett syndrome's vocalization, it is recommended to engage in a free improvisation, especially vocal improvisation and to leave ample space during the improvisation as well as during structured songs.

**Eye contact:**

Although several girls had their own unique form of communicating, there were many similarities between the girls’ informal communicative behaviors. The most obvious findings as far as eye contact is concerned are that all girls maintained eye contact throughout the preferred songs. They seemed extremely focused and intently concentrated while hearing the songs. It should be noted however that eye contact (although not as intense) was present during non-preferred songs as well. They never seemed to tire from this activity.

**Body movements:**

All girls moved their bodies during preferred songs. This may suggest their involvement in and the tuning to the music. The music provoked a rocking movement from side to side, back and forth and leaning forwards toward the researcher. This type of behavior is a natural one, especially in young children who fulfill classic criteria for diagnosis. Elisheva, Rachel and Meirav tapped their legs to the music and Tali also moved her head from side to side. The researcher was viewed this as similar to the rocking behavior presented by the rest of the participants. Aviv became very excited during preferred songs. At times she got out of her seat, began rocking and then returning to her seat, as if dancing. At times she put her hands on the researcher’s legs or put her leg on the researcher’s legs during singing. She seemed extremely content, focused and energized. This type of choosing activity and singing brought Aviv and the other participants to a high peak of excitement.

In the present study some of the girls moved rhythmically to the music. This was evident in rocking, head swaying, leg movement and hand movements. Burford & Trevarthen (1997) described that: “Girls with Rett syndrome can respond to repeated patterns of movements in
rhythmic/prosodic play to certain expressive forms transmitted in music” (p. 3). All girls, but Tali and Talia reacted at the end of the song by either leaning forwards or by looking away. Aviv leaned forward and tapped on the guitar at the end of the song. A few times when the researcher began coughing in the middle of the preferred song, Aviv stopped rocking, her face became serious, leaned forwards and began tapping on the guitar or the researcher's leg. When the researcher continued coughing, unable to continue singing, Aviv repeated this behavior. The researcher then continued singing and Aviv moved her body back into the seat, smiled broadly and lifted her leg on her seat. Other girls: Elisheva, Hilla, Meirav and Rachel looked away at the end of the song then looked back at the researcher. Tali stopped rocking and moving her head at the end of the song. From these informal behaviors, given the consistency with which they occurred, one can make a reasonable assumption that the participants either acknowledged the fact that the song was finished or signing a gesture wishing for ‘more’ singing. They may have wished for the singing to continue.

This behavior was very consistent and occurred after the song was sung and the girls were focusing on the researcher, smiling and rocking their bodies. This type of behavior was not present during non-preferred songs or during baseline. The informal behaviors demonstrated in the present study were referred to by Sigafoos et al. (1999). One out of the three girls in their study, leaned forwards when indicating ‘more’.

All girls leaned forward during the songs. It seemed as if they wanted to acknowledge the researcher by getting closer to her. Another possible assumption for moving towards the researcher might indicate a preference towards the song the same way as for turning the body towards a preferred musical piece, which has been found as indicator of musical preference in infants and toddlers (Trehub, 1985).

**Mouth movement:**

Three of the participants, Elisheva, Hilla and Rachel, opened and closed their mouth during preferred songs. This type of behavior may indicate some attempt or effort to sing, apparently mimicking the words. Hilla was observed during past individual music therapy to carry out
this type of mouth motion during songs and also during speaking interaction with the therapist. During these incidences the therapist left space or gave Hilla time to respond, which she did typically by speaking one to three, eligible words. The researcher has no knowledge if Elisheva or Rachel have making these mouth movements outside the research setting. Such behaviors were not observed by these three during baseline or when listening to non-preferred songs, marking this behavior as unique to uplifting/happy moments.

**Stereotyped hand movements:**

The incidence of hand movements in all participants (except Tali) was high during preferred songs probably due to the high arousal level and excitement evoked by these stimulating songs. The researcher tried several times to establish a direct link between tempo of hand movements and tempo of the songs. On many occasions the stereotyped hand movements matched the tempo of the preferred songs, which was typically during very fast tempo and quite energetic songs.

Hilla showed most accuracy of keeping the beat with her hands. This is interesting as although she rocked on several occasions, she moved much less than the others but almost always kept the beat to the music. Hilla has also been observed in the past (during group music therapy) to make hand motions during music in the ‘Monkey’ song (her favorite song). Tali’s hand movements were relatively less during the most preferred songs, but her body and head movements were much stronger and frequently were synchronous with the tempo of the music. It could be assumed that the girls are unable to maintain the tempo with more than one part of the body.

Since several of the girls maintained the beat of the music, it can be seen that the hands had an important part during music, as these were not just aimless hand movements. They appeared to have gained life and function during music.

During choice- making responses (throughout intervention and maintenance) their hands appeared more functional and developed into a more appropriate mean when indicating
choice. Several of the girls who initial chose through eye gaze began choosing by pointing with their face and thereafter began using their hand to touch the symbol. These findings coincides with Sigafoos et al. (1996) who noticed a trend in their subject of responding more functionally, rather than in an exploratory way, by using their hand to touch the symbol. This suggests that intervention procedures are effective in developing some functional requesting communications. It is important to encourage functional hand use, particularly related to independence skills such as choosing, or eating, and further studies aimed at promoting increases in hand use for the purpose of indicating choices are clearly necessary to consolidate the findings from this study.

**Behaviors typical to Rett syndrome - pathological behaviors as a group:**

In the present study hyperventilation was reduced when the girls were content while hearing the music. Wigram (1997) has, previously reported such reductions in hyperventilation when the girls are relaxed, during vibroacoustic therapy sessions. During baseline the participants might have been frustrated, which could explain the occurrence of hyperventilation. Woodyatt & Ozanne (1992, 1994) found that favorite songs increased hyperventilation and activity levels. In the present study, the girls increased hyperventilation while they were choosing the songs from the picture symbols. Once the chosen song was sung, the hyperventilation decreased and the activity levels (such as: hand movement, body movement, eye contact, vocalization) increased. It might imply that both types of behaviors exist in this population.

During baseline and non-preferred songs several behaviors were found in the present study to have been reduced (stereotyped hand movement and body movement). Wigram (1997) also found a reduction in bodily movements during vibroacoustic therapy when music was heard. The situation in both circumstances was too different to compare (sitting vs. lying down; ever changing vs. relaxing music; need to choose vs. no need to choose).

One argument could be that the girls (who employed these behaviors) in the present study did so not because they were relaxed, but as a result of ‘tuning out’, and so avoiding the situation.
The decrease in these behaviors is a positive one when the purpose was to provide relaxation (as was the case in Wigram’s 1997 study), but indicative of a lack of attention and interest in the present study. This is similar to Holdworth’s (1999) findings that the girls’ responsiveness decreased during unfamiliar music as compared to familiar music. Slow music (most of the present study’s less preferred songs were slow) was found both to relax, and to increase anxiety levels in girls with Rett syndrome (Mount et al., 2001). Although slower tempo was a contributor to less preference, one needs to take into consideration the total musical and non-musical features that may have affected the girls’ response.

**Summary:**

The participants in the present study, girls with Rett syndrome, demonstrated various emotional, communicative and pathological responses that might be interpreted as understandable messages. Some behaviors were frequent and exhibited by all participants, while others were unique and personal.

The researcher had undertaken no planned averaging of the behaviors of the participants that could be generalized to the wider populations. Nevertheless, the researcher’s knowledge of girls with Rett syndrome in general, and the participants in the present study in particular, provides support for some cautious interpretation. The detailed way in which the results have been analyzed, and anecdotal reports of other staff members has allowed the researcher to come to some fairly well-supported conclusions, based on these interpretations, that explain aspects of their behaviors.

The findings show that it is important to identify emotional reactions and different behaviors that can be interpreted as communicative attempts by a familiar figure of the client, such as a caregiver or a family member. Recognition of these behaviors and understanding their intended meaning will increase shared understanding. If these emotional and communicative attempts go unrecognized and unanswered, it may lead to the decay and demise of forms of behavior that, when recognized and interpreted, satisfy the needs of both client and caregiver.
There is also a danger that when a child’s needs and wishes go unrecognized withdrawal or more problematic behaviors might appear (Sigafoos et al. 2000).

Some of the following comments have been referred to in different sections of this thesis, however it was felt that summarizing limitations, generalizations, recommendations and clinical applications was warranted at the end of this chapter.
5.3 LIMITATIONS

The methods used in this study, the results of the trials and the conclusions that can be drawn are inevitably subject to certain limitations that need to be taken into account when considering the study within the wider context of research in the field. To begin with, it should be noted that the participants have previously been exposed to, and were engaged in activities where songs were offered for choosing. Each participant has had a different amount of experience and as a result the effects observed in the present study might be influenced by previous experience, thus enhancing the results. However, while this may have had a positive influence on the results, no previous attempts to evaluate choice making ability had been undertaken in a consistent way during intervention, and use of songs had been a part of a much more comprehensive intervention.

The interpretation of behaviors in participants as indicating emotional expression and communication was undertaken in this study, and certain behaviors were interpreted to have specific communicative meaning, and that the interpretation of such meaning is the responsibility of the adult. In the present study no direct verification from the participants in the research was possible due to their lack of verbal language and general cognitive impairments, and the researcher’s interpretation and categorization of these behaviors into communicative acts could be influenced by subjective judgment.

The researcher has had to rely on looking carefully and consistently, and identifying meaning from body language and facial expression. Nevertheless, detailed analysis has allowed this research to demonstrate enough frequency and consistency of response for those interpretations of emotional indicators and communicative behaviors to have a fair degree of content and criterion validity, and also to be reasonably reliable. The results of this study suggest that in future studies independent raters should be used to undertake a more detailed analysis of the frequency and duration of apparent communicative acts in order to achieve greater objectivity and internal validity. Interpreting non-verbal signals, body language and gesture in attaching precise emotional meaning that can be interpreted consistently is nevertheless a complex process with any population that is difficult to validate.
5.4 GENERALIZATION

This study did not involve the use of a randomized control design. The participants acted as their own controls. The seven participants engaged in this study serve as case examples, indicative of capacities that girls with Rett syndrome may have.

The study showed that the participants responded well to the musical interventions presented to them. It may be expected any future controlled studies will demonstrate similar findings generalizable to the wider population.

The methodology that was used has been found appropriate in this Rett syndrome group and could be applied more widely to studies that target populations with a similar level of disability.

Importantly, this study showed that the participants expanded their communication skills into other areas of daily living. They were able to use picture symbols as an aid to communication and choice making during interactive story telling, mealtimes, using computer games, and generally in the classroom, as well as with their caregivers in their homes. The success of this transfer of learned communicative abilities enables us to reject assumptions by Budden et al. (1990), who report that although girls with Rett syndrome are able to initiate some form of communication in different settings, they cannot generalize this behavior.

The songs that were chosen for this research were shown to attract interest and response from this population and could be used with other developmentally disabled population because of their simplicity, repetitiveness, predictable structure and attractive melodies. In this study the 18 songs (familiar and unfamiliar) were randomly assigned, allowing for potential positive and/or negative response to each of the songs. The consistent responses found both within and between participants to randomized presentation strengthen the findings of the present study.
5.5 RECOMMENDATION FOR FURTHER RESEARCH

The findings from this study show that the participants in this study could demonstrate intentional choice making ability, and further investigation is warranted to determine whether these very positive results would generalize to the wider population. It is also apparent from the findings that these abilities are improving over time with constant support, and such support is vital.

Children with Rett syndrome need more time to learn new procedures before achieving their potential. This might mean that with every intervention employed with this population, a preliminary intensive acclimatization period is needed (in this study three times per week over a 2-3 weeks period) before the achievement of a basic skill level for learning. The success of this study may be partially due to the previous experiences the participants had with the researcher (in clinical sessions) and the setting. It is this researcher’s recommendation that future investigation will take place in known situations and with a person with whom the participants feel comfortable and familiar. While baseline orientation and familiarity may result in the use of participants who are not ‘naive’, the argument in favor of such a ‘prepared’ subject in this particular population is that they take time to adjust to new stimuli, and respond at their optimum level. Comparison between initial baseline data, and data collected during intervention will, in any case, demonstrate any significant differences if they are to be found. The relationship between the therapist and the girls is also considered important as the basis of a successful intervention (Wigram, 1991, 1995; Hill, 1997), and in the case of this study, this aspect has undoubtedly played a part in achieving positive results. It is hypothesized that implementation of research design not answering to this primary stage might yield different or less powerful results. This needs further investigation.

Due to the results achieved in this investigation, maintenance sessions are recommended for future investigations when learning process is evaluated in a non-verbal multi-handicapped population, thus enabling the child to enhance and internalize learned experiences, stimuli and procedures.
It is this researcher’s basic belief that improved skills can only be displayed as such, when change is detected within the scope of the child’s daily activities. In order to achieve such changes, and convey the treatment room developments and achievements to real life, many opportunities of choice making should be available to the girls throughout the day within and across daily activities and future research should be expanded into these areas. All participants in the present study established fluency in choice making as a result of ample opportunities and this should be taken into consideration. It is important to maintain the skill by generalizing it to other places. The choice opportunities should be “highly individualized and carefully planned to ensure that simple, but meaningful opportunities are not overlooked” (Bambara et al., 1995, p. 186). Most opportunities can be formed into an occasion for choice. It is recommended for individuals with Rett syndrome to use stimuli with high motivational value, such as music, as Sigafoos et al. (1996) found in their study when teaching girls with Rett syndrome to request preferred objects.

It is suggested from this study that specific elements in the music have certain emotional, communicative and pathological effects on this population. Certain types of behaviors and musical elements are worth monitoring. Future research could investigate musical elements as well as emotional, communicative and pathological responses. Facial expression, bodily movement (for example leaning forward), anticipation, and other responses noted indicate some important and powerful effects of some elements of music that influence this population, and this may establish a deeper understanding of non-verbal communicative acts.
5.6 CLINICAL APPLICABILITY

The results of this study revealed that songs in music therapy are a powerful medium for girls with Rett syndrome. This should be taken into consideration by both clinicians in different disciplines, and teachers/educators working with this population, especially music therapists and music teachers.

Specific individual song preferences were found in this study. Establishing each child’s individual preference is an important first step when trying to build a proper motivational factors to achieve the cooperation of the child in any activity calling for active participation. Although the girls might reveal that they sustain their preferences for a period of time, these preferences could change.

When determining what type of songs to use during music therapy intervention it will be more effective if they encompass some of the musical elements found in this research (always remembering each child’s personal preference and musical background can and will differ).

The presentation (singing) of the songs should address the expressive part of the music and at the same time needs to be presented as truly as possible to the musical structure and its style. Being tuned to the child’s emotions and reflecting these emotions musically does result in a meaningful form of interaction between therapist and the child. This type of musical/emotional interaction establishes an important dynamic foundation for growth and learning in the child with developmental disability in general and with Rett syndrome in particular.

The participants in this study revealed various types of responses when using certain songs and musical elements. Clinicians should look for these different musical elements (discussed in Chapter 4) and how they may effect their patients with Rett syndrome. One can look at ways that the girls express communication intentionally, some of the ways they express emotions and the differentiation of those things from the pathological behaviors.

In this study the girls vocalized more in between the songs and at the end of the sessions. The songs may have provoked vocalization, but they vocalized less than was hypothesized during
singing. In order to enhance vocalization it is recommended to give ample space and time for
the girl to respond either in pre-composed songs or in improvised activities.

Age appropriate songs are important when selecting songs for this population. The older girls
in this study only rarely chose songs that they liked when they were younger (which were also
slower, gentler and more in the nature of lullabies than the preferred songs). In this study they
chose the more compound songs. The level of the songs should be taken into consideration
when working with this population, however the expressive aspect when performing should
always be an integral part of the songs.

The form in which the girls indicated their choice changed over time. A few of the girls in this
study began choosing by scanning the symbols then starring and focusing on one symbol. As
time progressed they began choosing by pointing with their face and nose and further along
several of the girls began indicating their choice by touching the symbols with their hands. It
seems that the more motivated and secure they felt, the more changes they made in the form of
communication. This demonstrates that the girls may be able to perform at a higher level than
initially assumed based on what they reveal. It is therefore important for the clinician to trust
the girls and give them ample time for growth. It also implies that the girls can use their hands
when motivated by holding down gently the non-dominant hand while freeing the dominant

This study found that all girls initially had long elapse times prior to responding to the
stimulus. The reduction in response time is believed to result from proper and meaningful
interest that was achieved when a strong motivational factor, such as music (songs) was used.
The stimulus in this research (songs) was just such a motivation factor and the girls seemed
eager to respond to the stimulus. It is vital to allow the girls ample time to respond, especially
when engaged in a new activity. It is also important to give the direction needed and wait
silently and patiently for the girl to respond. With practice and interest her response will
become faster and consistent. When participants in a study like the present one, establish
fluency in choice making, it is important to maintain the skill by generalizing it to other areas
than the therapy room.
CONCLUSION

The present research study revealed hidden abilities and skills in a population perceived as non-educable until not long ago. Participants in this study, girls with Rett syndrome were found to be very responsive to song singing of composed songs. They demonstrated capacity to choose intentionally and established fluency in choice making as their response time was reduced dramatically. The participants were learning throughout the study and sustained learning over time as they seemed to have internalized what they had learned. The girls with Rett syndrome included in this study were able to express clear song preferences. Musical and non-musical elements were found to influence whether or not a participant preferred one song to another, which warrants further generalization to providing them with the opportunities to express their needs and wishes and gain/sustaining control over their immediate environment.

In this study, girls with Rett syndrome demonstrated a wide variety of emotional and communicative behaviors. The girls showed ‘positive’ emotions (smile, laughter, shout) and behaviors (eye contact, rocking, leaning forward and more) when hearing songs, most obviously when hearing preferred songs, and reacted negatively (i.e. frown face, cry, head turn, or leave seat) when they received no songs in response to making a choice. The intensity of stereotypic hand movement changed according to the music, and several girls kept rhythm to the music. All girls demonstrated consistency of response at certain places in the songs. They all smiled or laughed at almost the exact same place of a song, and at times anticipated these places in the song, responding early. These places would typically have an interesting sound, pitch or rhythm.

I have been working with girls with Rett syndrome for the past 13 years. During this period the hidden skills of this population in learning and communicating as well as their profound attraction to music became clear to me. Clinical experience has shown me that once children with Rett syndrome find an interesting and motivating environment, such as music therapy, they are likely to become very involved, attuned and more likely to learn. It is therefore natural that music should be utilized as a medium for promoting communication for individuals with Rett syndrome. As a result, this study was set forth.
The process of this study was extremely satisfying and rewarding as it provided important answers to the initial hypothesis and sub-questions of this research. It demonstrated that girls with Rett syndrome revealed their potential through songs in music therapy, but this might only be the beginning of what may be hidden behind this debilitating disorder. This form of intervention, songs in music therapy seems to be an important basis for their development and growth and it is worth exploring and investigating it further by music therapists working with this population.
SUMMARY

Theoretical background

Rett syndrome is a severe neurological disorder affecting mainly females and is considered to be the most common cause of multiple-disability among them. Individuals with Rett syndrome suffer from cognitive impairment, placing them within the range of severe to profound learning disability. One of the main features of this population is a severe impairment of receptive and expressive communication, demonstrated in the majority of cases in a lack of verbal language which either never develops, or which is lost during the regressive phase of the syndrome.

Females with Rett syndrome are reported to be responsive to music. Consequently, music therapy has been indicated as a relevant treatment where practitioners have demonstrated that intervention promotes and motivates their desire to interact and communicate, as well as to stimulate many aspects of development including: choice making, enhancing vocalization, improving eye contact, and opening channels for emotional and communicative expression.

Aim and approach

The purpose of this research was to investigate the following question:

**Can songs in music therapy enhance communication in girls with Rett syndrome?**

The sub questions supporting the primary research question were?

1. Are girls with Rett syndrome able to make intentional choices?
2. Are girls with Rett syndrome able to learn and sustain learning over time?
3. Do girls with Rett syndrome reveal consistent preferences through choices they make?
4. How do girls with Rett syndrome demonstrate emotional and communicative behaviors?
Response time was also measured during analysis of the data initially not part of the sub-questions. Data it will be referred to in the result section.

Seven girls with Rett syndrome, ages 4-10 participated in this study. A single case, multiple probe design was used involving 30-minute trials, three times per week for a period of eight months, during which 18 familiar and unfamiliar children songs were presented to the girls. The songs were grouped into three sets of songs, six in each set, and the participants were offered the chance to choose their preferred songs, following which analysis was made of their responses.

Baseline measurements for about 3-4 sessions were employed where songs picture symbols or orthographies were exposed to each participant while they were asked to choose a song. The songs were not sung during the baseline period whether or not they chose the song. The purpose of the baseline was to determine consistency and intentionality in the participant’s abilities to make choices.

After a stable baseline had been established, intervention began and the song was sung to a participant after she had made her choice. Post intervention maintenance probe sessions were scheduled in order to determine whether the participants had internalized learning and sustained choice making skills. These were undertaken at fixed time intervals after the participants’ completion of the trials, given that they had met pre-determined criterion to establish reliability of choice making. All seven participants met this criterion.

The entire baseline, intervention and maintenance trials in the study were video taped and then analyzed. All data was viewed for inter-observer reliability both for dependent and independent reliability. To ensure inter-observer reliability, 20% of the data was randomly selected, observed, and scored by an independent observer. This revealed 96% inter-observer agreement with the researcher for scoring of the dependent variables, and 99% inter-observer agreement for scoring the independent variable.
Effect size calculations were undertaken to evaluate the potential effects of intervention compared with baseline for all participants in the study, and descriptive statistical analysis was applied to illustrate the results achieved by the participants in choice making.

**Results and Discussion**

**Results and Discussion of the research methodology:**
The present study found the multiple probe design (a research tool not frequently used in music therapy) to be most appropriate in collecting and systematically analyzing effects over time, and determining the ability of the participants to acquire and sustain choice making skills. This design has been found sensitive enough to differentiate individual abilities and variables and was especially suited for evaluating whether ability was sustained following periods of no intervention.

**Results & Discussion will be presented according to the research questions:**

- **Girls with Rett syndrome are able to make intentional choices.**
The study found evidence on a single case basis that girls with Rett syndrome in this study, revealed a strong motivation to choose songs, and confirm their choice, thus demonstrating intentional choice making.

These findings suggest that the songs used in this study were effective in revealing potentials for intentional choice making, demonstrating through the process evidence of clear preferences.

The participants are from a clinical population that are predominately thought of as uneducable, with severe learning disability and who are considered to be pre-intentional in their communication.
- **Girls with Rett syndrome reduce response time over time.**
  Measuring response time was undertaken during the trials, as an additional indicator of intentionality, considering that girls from this clinical population are considered to have physiologically delayed responses. Initially the participants responded to choices of songs with a delay response of 15-45 seconds. Within three to four sessions all participants responded within or less than 15 seconds. The reduction of response time implies motivation for the songs presented, and the rapid reduction in response time requires further investigation and explanation.

- **Girls with Rett syndrome are able to learn and to sustain learning over time.**
  All participants demonstrated their ability to learn, and made impressive progress in learning within and between the sets. The participants sustained learning over time and seemed to have internalized what they had learned, as they kept their consistency of choosing scores at a high level (averaging at 97.5%).

  This occurred despite the fact that there was a three-month interval between the end of the intervention and the last maintenance session. The overall mean effect size for all the participants’ choosing abilities comparing baseline with intervention is $d = 8.39$, and baseline with maintenance is $d =10.64$, clearly indicating a very large effect of intervention and maintenance over baseline in the participants ability to demonstrate choice making.

- **Girls with Rett syndrome reveal consistent preferences through choices**
  It can be concluded from the results, that the participants in this study are able to choose and have song preferences. The participants had the same five most and five least preferred songs, but each had clearly individual song preferences. Not only did they choose certain songs more than others did, but they also demonstrated their preference through emotional and communicative behaviors. Their responses were
very different when the most preferred and when the least preferred songs were sung to them.

Musical and non-musical elements were found to influence whether or not a participant preferred one song to another. Familiarity was a strong indication and all girls preferred familiar songs to unfamiliar ones. The results indicate that the participants had the ability to differentiate between the songs.

- **Girls with Rett syndrome demonstrate emotional and communicative behaviors**

  The girls in this study reacted negatively (i.e. frown face, cry, head turn, or leave seat) when they received no songs in response to making a choice. They showed ‘positive’ emotions (smile, laughter, shout) and behavior (eye contact, rocking, leaning forward and more) when hearing songs, most obviously when hearing preferred songs, supporting the argument that choices were made on the basis of likes and dislikes.

  All girls demonstrated consistency of response at certain places in the songs. They all smiled or laughed at almost the exact same place of a song, and at times anticipated these places in the song, responding early. These places would typically have an interesting sound, pitch or rhythm. The findings also show that their response tailed off over time after the songs had been presented many times. Other findings revealed that most participants presented high levels of hand movements when being sung to. This finding corresponds well with cases described by other researchers and clinicians in the field.

  Vocalization increased in quantity and intensity after the song had been sung, and mostly at the end of the sessions. The findings also suggest that in order to elicit or enhance their vocalization, it is recommended to engage in a free improvisation or leave spaces for response during structured songs. The results also reported reductions in hyperventilation in some participants when they became content and relaxed when the girls were content and relaxed while listening to the songs.
The participants in the present study demonstrated a variety behaviors and emotional responses that can be translated into understandable messages by attentive caregivers and therapists.

**Results and discussion of the musical analysis:**

Eighteen children’s songs were used in the present study. The songs were short songs with one verse and were typically repeated three times. Some musical elements such as: fast tempo, tempo variability (accelerandos, ritardandos, pauses and others) vocal play and vocal sounds, interesting rhythmical and melodic patterns were found to be attractive to the participants (according to the analysis of emotional responses) and appeared to increase their desire to become active learners. All participants seemed to appreciate some specific musical elements, as there were consistent responses across subjects at the same places in the music. These factors should be taken into consideration when working with this population.

**Summary**

Participants in this study have been found to be very responsive to song singing a technique used in music therapy that involves the use of composed songs. The capacity to choose, demonstrated in this study, needs to be taken into consideration in assessment and evaluation of this population when determining their everyday needs and learning potential. If the girls with Rett syndrome included in this study are able to demonstrate a capacity for learned choice making and the expression of preferences, this warrants further generalization to providing them with the opportunities to express their needs and wishes. Choice making is a prerequisite for improving quality of life, and gain/sustaining control over one’s immediate environment.

As a result of this study the participants expanded their communication skills into other areas of daily living. These included: picture symbols during interactive storytelling, during mealtime, computer games in the classrooms as well as with their caregivers at home. When working with the population of learning disabilities to enhance the use of spontaneous communication, one should arrange for plenty of opportunities set up throughout the day and within different situations.
When participants in a study like the present one, establish fluency in choice making, it is important to maintain the skill by generalizing it to other areas than the therapy room. The present research revealed hidden abilities and skills in a population perceived as non-educable until not long ago.

**Limitations**

The present results have been achieved by obtaining data from a relatively small group of participants, (N=7). It was a single subject design, so there was no control to compare. The control established was within subjects – where the baseline period acted as a pre-intervention control. No comparison was made with another form of intervention, and the results from the seven participants can not be generalized to the wider population. Implications can be drawn that support the need for further research with a larger sample in order to achieve replication of these results.
SUMMARY (DANISH)

Resumé

Forøget kommunikation hos piger med Rett syndromet gennem sange i musikterapi

af Cochavit Elefant

Teoretisk baggrund

Rett syndromet er en alvorlig neurologisk sygdom, der kun rammer kvinder og som betragtes som værende den mest almindelige grund til multipel-handicap hos dem.

Individer med Rett syndromet lider af kognitiv svækelse, og dette anbringer dem inden for rammerne af alvorlig til dybtgående indlæringsevne. Én af hovedtrækkene for denne gruppe er en alvorlig svækelse af receptiv og ekspressiv kommunikation, der i hovedparten af tilfældene viser sig i en mangel på verbalt sprog, som enten aldrig udvikler sig eller forsvinder i den regressive fase af syndromet.

Kvinder med Rett Syndromet rapporteres at være modtagelige over for musik. Derfor er musikterapi blevet nævnt som en relevant behandling, hvor udøvere har demonstreret deres ønske om at interagere og kommunikere så vel som at stimulere mange udviklingssider: udvælgelsesevne, forbedring af vokalisering, forbedring af øjenkontakt og åbningskanaler for følelsesmæssig og kommunikativ udtryksevne.

Mål og fremgangsmåde

Hensigten med denne forskning var at undersøge følgende spørgsmål:
Underspørgsmålene, der understøtter det primære forskningsemne, var:
Kan sange i musikterapi forbedre kommunikationsevnen for piger med Rett syndromet?

1. Er piger med Rett syndromet i stand til at foretage bevidste valg?
2. Er piger med Rett syndromet i stand til at lære og bibeholde det indlærte efter en periode?
3. Afslører piger med Rett syndromet grupperede præferencer gennem de valg, de foretager?
4. Hvordan viser piger med Rett syndromet følelsesmæssig og kommunikativ opførsel?

Svartiden blev også målt ved hjælp af analyse af data, der ikke i begyndelsen var en del af undersøgelsesmålene.

7 piger med Rett syndromet, fra 4-10 år, deltog i denne undersøgelse. Et multiple undersøgelsesudformning (enkelt tilfælde) blev benyttet, idet den involverede 30-minutters forsøg tre gange om ugen i en periode på 8 måneder, hvor 18 kendte og ukendte børnesange blev præsenteret for pigerne. Sangene blev grupperet i 3 sæt med 6 i hver, og deltagerne fik tilbudt at vælge deres favoritsange, hvorefter der blev udfærdiget en analyse af deres svar.

Grundniveaumålere blev benyttet for omkring 3-4 sessioner, hvor sangbilledsymboler eller ortografier blev vist til hver deltagere, mens de blev bedt om at vælge en sang. Sangene blev ikke sunget i løbet af grundperioden, hvad enten de valgte sangen eller ej. Hensigten med grundperioden var at fastlægge overensstemmelse og vilje hos deltagere til at foretage valg.

Efter at en fast måling var blevet etableret, begyndte interventionen, og sangen blev sunget for deltageren, efter at denne havde foretaget sit valg. Sessioner med efterfølgende interventionsbibeholdelsesprøve blev planlagt for at afgøre, om deltagerne havde internaliseret indlæring og gennemført vedvarende udvælgelsesevner. Disse blev foretaget med faste tidsintervaller, efter at deltagere var færdige med forsøgene, forudsat at de havde klaret forudsatte kriterier til etablering af pålidelige udvælgelsesevner. Alle syv deltagere klarede dette kriterium.
Alle grundforsøgene, interventions- og opretholdelsesforsøgene i dette forsøg, blev videooptaget og så analyseret. Alle data blev gennemset for interobserverpålidelighed, både afhængig og uafhængig pålidelighed. For at sikre interobservationspålideligheden var der en uafhængig observator, der observerede og førte regnskab med 20% af data, der var valgt tilfældigt. Dette afslørede 96%'s interobservatørenighed med forskeren i bedømmelsen af de afhængige variable, og 99%'s interobservatørenighed i bedømmelsen af de uafhængige variable.

Effektstørrelseskalkulationer blev foretaget for at evaluere de potentielle effekter af interventionen sammenlignet med grundniveauet for alle deltagerne i forsøget, og deskriptiv statistisk analyse blev gennemført for at illustrere de udvælgelsesresultater, der blev opnået af deltagerne.

Resultater og diskussion.

Forskningsmetodens resultater og diskussioner:
Det nuværende forsøg vurderede multiple forsøget (et forskningsredskab, der ikke ofte bruges inden for musikterapi) til at være det mest passende til at samle og systematisk analysere effekter over tid, og til at afgøre deltagernes evne til at opnå og bibeholde udvælgelsesevner. Dette design er blevet bedømt til at være sensitivt nok til at differentiere individuelle evner og variable og var specielt egnet til en evaluering af, om evner blev bibeholdt, når der var perioder uden intervention.

Resultater og diskussion vil blive præsenteret ifølge forskningsspørgsmålene:

Piger med Rett syndromet er i stand til at foretage bevidste valg.
Forsøget fandt tegn i et enkelt forsøg på, at piger med Rett syndromet afslørede en stærk motivation til at vælge sange og at bekræfte deres valg og således viste bevidst udvælgelse. Disse resultater viser, at sangene, der blev brugt i forsøget, var effektive til at afsløre evner for bevidst udvælgelse, idet de gennem processen viste bevis på klare præferencer. Deltagere er fra en klinisk befolkningsgruppe, som mest opfattes som mennesker, der ikke kan lære noget,
med alvorlig mangel på indlæringsevne, og som betragtes som værende ubevidste i deres kommunikation.

**Piger med Rette syndromet reducerer respons med tiden**


**Piger med Rett syndromet er i stand til at lære og at opretholde det indlærte efter en periode.**

Alle deltagerne viste deres evne til at lære og gjorde imponerende indlæringsfremskridt inden for og mellem forsøgene.

Deltagerne opretholdt indlæringen efter noget tid og så ud til at have internaliseret, hvad de havde lært, samtidigt med at de beholdt deres evne til at vælge point på et højt niveau (gennemsnit på 97,5%). Dette skete, på trods af det faktum, at der var et tremåneders interval mellem slutningen af interventionen og den sidst afholdte session. Den samlede gennemsnitlige effektstørrelse for alle deltagernes udvælgelsesevne, når man sammenlignede grundlinien med interventionen, er \( d = 8,39 \), og grundlinien med vedligeholdelsen er \( d = 10,64 \), hvilket klart viser en meget stor effekt af intervention og vedligeholdelse over grundniveauet i deltagernes evne til at vise udvælgelsesevne.

**Piger med Rett syndromet afslører vedholdende præferencer gennem udvælgelse**

På baggrund af resultaterne kan det konkluderes, at deltagerne i dette forsøg er i stand til at vælge og have præferencer for sange. Deltagerne har de samme 5 mest og de samme 5 mindst foretrukne sange, men alle har klart individuelle sangpræferencer. De valgte ikke kun bestemt

**Piger med Rett syndromet viser emotionel og kommunikativ opførsel**

Pigerne i forsøget reagerede negativt (dvs. rynkede panden, dregede hovedet eller forlod stolen), når de ikke modtog sange som et svar på at have foretaget et valg. De viste 'positive' følelser (smil, latter, råb) og opførsel (øjenkontakt, røkkede frem og tilbage, lænede sig fremover og flere ting), når de hørte sange, mest tydeligt når de hørte foretrukne sange, hvilket understøtter det argument, at valgene var foretaget på baggrund af noget, de kunne lide og ikke lide.


Vokaliseringen steg i kvantitet og intensitet, **efter** at sangen var blevet sunget, og mest i slutningen af sessionerne. Resultaterne anvider også, at for at lokke deres vokaliserings frem, anbefales det at gå ind i en fri improvisation eller at efterlade plads til svar i løbet af strukturerede sange. Resultaterne rapporterer også reduktion i hyperventilation hos nogle deltagere, når pigerne blev tilfredse og afslappede, mens de lyttede til sangene. Deltagerne i det nuværende forsøg viste forskellig opførsel og emotionel respons, som kan oversættes til forståelige meddelelser af lyttende plejepersonale og terapeuter.
Resultaterne og diskussion af musikalsk analyse

18 børnesange blev brugt i det nuværende forsøg. Sangene var korte sange med ét vers og blev typisk gentaget tre gange. Nogle musikalske elementer så som: hurtigt tempo, temposkift (accelerandos, ritardandos, pauser og andre), vokalt spil og vokale lyde, interessante rytmiske og melodiske mønstre var tydeligt attraktive for deltagerne (ifølge analysen af de emotionelle svar) og viste at øge deres lyst til at blive aktive indlæringsdeltagere. Alle deltagerne så ud til at kunne lide nogle specielle musikalske elementer, lige så vel som der var vedholdende svar på temaer de samme steder i musikken. Disse faktorer bør tages i betragtning, når der arbejdes med denne persongruppe.

Resumé

Deltagerne i dette forsøg har vist sig at være meget responderende over for sange, der blev sunget, en teknik, der brugt i musikterapi involverer brugen af komponerede sange. Evnen til at vælge, vist i dette forsøg, bør tages i betragtning i vurderingen og evalueringen af denne persongruppe, når man afgør deres behov og indlæringsevne i hverdagen. Hvis pigerne med Rett syndromet, der er involveret i dette forsøg, er i stand til at demonstrere en evne til at lære at udvælge og udtrykke præferencer, garanterer en yderligere generalisering at skaffe dem en mulighed for at udtrykke deres behov og ønsker. Udvælgelse er en forudsætning for at forbedre livskvaliteten og at skaffe kontrol over sine nære omgivelser.

**Begrænsninger.**

De nuværende resultater er opnået ved at samle data fra en relativ lille gruppe deltagere (N = 7). Det var et enkelt forsøg, så der var ikke nogen kontrolgruppe at sammenligne med. Den etablerede kontrol var inden for temaer - hvor grundniveauet blev opfattet som en kontrol før interventionsperioden. Der blev ikke lavet nogen sammenligning med en anden form for intervention, og resultaterne fra de 7 deltagere er ikke generelle for en større persongruppe. Der kan ses implikationer, som støtter behovet for yderligere forskning med et større antal mennesker for at opnå kontrol af disse resultater.
Appendix I
Parental consent

Dear Mr. & Mrs._______________

I will be conducting a research project as part of my Ph.D. studies in Music Therapy. The topic of my research is: "Enhancing Communication Skills of Girls with Rett Syndrome through Songs in Music Therapy". The study will focus on the effect music therapy has on communication skills with this population. In order to undergo this study I need your permission to have your daughter participate in a music therapy program as a member in this study. This letter is meant to explain the procedures of the study.

The research will begin at the end of January 1999 and will continue until the end of this school year or into the fall. Each girl will receive individual half an hour sessions of music therapy three times per week which will be conducted by myself in the music therapy room at Beit Issie Shapiro. During the music therapy sessions, your daughter will make choices of familiar and unfamiliar songs through picture and/or orthographic symbols. Once a choice of a song has been made, the song will be sung to her.

All sessions will be videotaped and then analyzed looking at different elements such as intentional communication: choice making of songs, vocalization, smiles, laughs, eye contact and other body gestures. I will also look at the amount of time it takes for each girl to make a choice and at the duration and intensity of her hand stereotypic movements during the sessions. The videotapes will be viewed by other professionals for coding purposes only. The coders will not have access to any personal data regarding your child.

My music therapy supervisor at the University of Aalborg in Denmark (where my official studies take place) is Prof. Tony Wigram who is a leading figure in music therapy with Rett Syndrome. He has conducted several research projects, written many articles and books on this population. My advisor in the study in Israel is Dr. Orit Hetzroni from the University of Haifa who is a familiar figure to the girls and
conducted research at Beit Issie Shapiro. She will advice me throughout the project. In addition, Dr. Dana Roth, research and development director at Beit Issie Shapiro will be monitoring and supporting the project. Beit Issie Shapiro supports this study which hopefully will promote our understanding and develop better treatments for girls with Rett Syndrome.

I hope you can value the need for such a research for your daughter's benefit and as a contribution to the Rett Syndrome field as a whole, and agree to have your daughter participate by signing and returning the attached permission slip.

Please call me at 09-767-8588 if you would like to receive any additional information or if you may have any questions concerning this research.

Thanking you for your cooperation.
Sincerely,
Cochavit Elefant
Music Therapist
Beit Issie Shapiro

I permit __________________ to participate in the music therapy name research and allow to use videotape material for the purpose of learning.
I do not permit __________________ to participate in the music therapy name research.
I wish for my daughter's name to be changed during presentations yes/no

__________________
Parents Name

__________________
Date

__________________
Parents Signature

copy: Dr. Dana Roth, research and development dept., Beit Issie Shapiro.
# Appendix II

## General Recording Form

<table>
<thead>
<tr>
<th>Date</th>
<th>Session #</th>
<th>Set #</th>
<th>Sequence in set</th>
<th>Probe</th>
<th>Maintenance</th>
<th>Comments</th>
</tr>
</thead>
</table>


## Appendix III

### Direct/Video Data Collection Form

**Name:**

**Duration of session:**

**Date:**

<table>
<thead>
<tr>
<th>Song</th>
<th>Total Song Selection</th>
</tr>
</thead>
<tbody>
<tr>
<td>----</td>
<td></td>
</tr>
<tr>
<td>Indicates and repeats song preference</td>
<td>1</td>
</tr>
<tr>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>4</td>
<td>4</td>
</tr>
<tr>
<td>5</td>
<td>5</td>
</tr>
<tr>
<td>Form of selection (hand, nose, chin, eye, gaze)</td>
<td></td>
</tr>
<tr>
<td>Time between stimulus and response</td>
<td></td>
</tr>
<tr>
<td>Smiles, laughs or shouts when stimulus presented</td>
<td></td>
</tr>
<tr>
<td>Eye contact when stimulus presented</td>
<td></td>
</tr>
<tr>
<td>Cries when stimulus presented</td>
<td></td>
</tr>
<tr>
<td>Stereotypic hand movement</td>
<td></td>
</tr>
<tr>
<td>Vocalization when making choice</td>
<td></td>
</tr>
<tr>
<td>Detached sound utterances</td>
<td></td>
</tr>
<tr>
<td>Sustained sound utterances</td>
<td></td>
</tr>
<tr>
<td>Uses inflection</td>
<td></td>
</tr>
<tr>
<td>Uses proto words</td>
<td></td>
</tr>
<tr>
<td>Uses words</td>
<td></td>
</tr>
<tr>
<td>Body movement</td>
<td></td>
</tr>
<tr>
<td>Mouth open/close</td>
<td></td>
</tr>
<tr>
<td>Teeth grinding</td>
<td></td>
</tr>
<tr>
<td>Hyper ventilation</td>
<td></td>
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</table>
## Appendix IV

### Behavior Analysis Form

<table>
<thead>
<tr>
<th>Form of selection (hand, nose, chin, eye, gaze)</th>
<th>Song 1</th>
<th>Song 2</th>
<th>Song 3</th>
<th>Song 4</th>
<th>Song 5</th>
<th>Song 6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Indicates song preference</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td></td>
<td>3</td>
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</tbody>
</table>

| Time between stimulus and response            |        |        |        |        |        |        |
| Smiles, laughs or shouts when stimulus       |        |        |        |        |        |        |
| presented                                    |        |        |        |        |        |        |
| Eye contact when stimulus presented          |        |        |        |        |        |        |
| Cries when stimulus presented                |        |        |        |        |        |        |
| Stereotypic hand movement                    |        |        |        |        |        |        |
| Vocalization when making choice              |        |        |        |        |        |        |
| Detached sound utterances                    |        |        |        |        |        |        |
| Sustained sound utterances                   |        |        |        |        |        |        |
| Uses inflection                              |        |        |        |        |        |        |
| Uses proto words                             |        |        |        |        |        |        |
| Uses words                                   |        |        |        |        |        |        |
## Appendix V
### Communication, Emotional and Pathological Observation Form

Starting time of session: \(\text{________} \)  
End time of session: \(\text{________} \)  
Session: \(\text{________} \)  
Duration of session: \(\text{________} \)  
Date: \(\text{________} \)

<table>
<thead>
<tr>
<th>Frequency</th>
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<th>Frequency</th>
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<tbody>
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<td>Song 3</td>
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<td>Song 6</td>
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</tbody>
</table>
## Appendix VI

### Duration of Vocalization, Emotional and Pathological Behavior Form

<table>
<thead>
<tr>
<th>Duration of Vocalization, Emotional and Pathological Behavior Form</th>
</tr>
</thead>
<tbody>
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<td>Starting time of session: _______________________________</td>
</tr>
<tr>
<td>End time of session: _______________________________</td>
</tr>
<tr>
<td>Duration of session: _______________________________</td>
</tr>
<tr>
<td>Name: _______________________________</td>
</tr>
<tr>
<td>Session: _______________________________</td>
</tr>
<tr>
<td>Date: _______________________________</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th></th>
<th>Song 1</th>
<th>Song 2</th>
<th>Song 3</th>
<th>Song 4</th>
<th>Song 5</th>
<th>Song 6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Smile, laughs or shouts during singing</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cries during singing</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Eye contact during singing</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Moves body during singing (rocking)</td>
<td></td>
<td></td>
<td></td>
<td></td>
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<td></td>
</tr>
<tr>
<td>Moves body to the rhythm of music</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Stereotypic hand movement</td>
<td></td>
<td></td>
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<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Stereotypic hand movement to the rhythm of music</td>
<td></td>
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<td></td>
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<td></td>
</tr>
<tr>
<td>Moves legs during singing</td>
<td></td>
<td></td>
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</tr>
<tr>
<td>Moves legs to the rhythm of the music</td>
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<tr>
<td>Loud vocalization</td>
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<tr>
<td>Soft vocalization</td>
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<tr>
<td>Uses proto-words</td>
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</tr>
<tr>
<td>Smile, laughs or shouts at the end of song</td>
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</table>
# Appendix VII

## Song Selection Frequency Form

<table>
<thead>
<tr>
<th>Date</th>
<th>Monkey</th>
<th>Crocodile</th>
<th>Farm</th>
<th>Hands</th>
<th>Bird</th>
<th>Turtle</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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</tbody>
</table>

**Set 1**

<table>
<thead>
<tr>
<th>Tree</th>
<th>Spider</th>
<th>Rabbit</th>
<th>Bee</th>
<th>Frog</th>
<th>Star</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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</tbody>
</table>

**Set 2**

<table>
<thead>
<tr>
<th>Elephant</th>
<th>Butterfly</th>
<th>Train</th>
<th>Nut</th>
<th>Chick</th>
<th>Fish</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
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</tbody>
</table>
Spider

Allegretto

A

mf
deep voice
high pitch

Bee

Allegro

D

mf
A7
D a tempo
rit.
glass.
ad lib. A7

(oops) (buzz)
Train

Allegro \( \frac{4}{4} \) \( \frac{4}{4} \)

\( \text{mf} \)

\( A \)

\( A \)

\( E7 \)

\( A \)

\( A \)

\( D \)

\( E7 \)

\( A \)

\( E7 \)

\( A \)

\( A \)

\( E7 \)

\( A \)

\( accel \)

\( E7 \)

\( A \)

\( rit \)
Farm

Allegro \( \frac{d}{= \frac{32}{32}} \)

\[
\begin{array}{c}
\begin{array}{c}
G7 \quad C \\
Am \quad Dm \quad G7 \quad C
\end{array}
\end{array}
\]

\[
\begin{array}{c}
\begin{array}{c}
G7 \\
C
\end{array}
\end{array}
\]

\[
\begin{array}{c}
\begin{array}{c}
Am \quad Dm \\
G7 \quad C
\end{array}
\end{array}
\]

\[
\begin{array}{c}
\begin{array}{c}
G7 \\
C
\end{array}
\end{array}
\]

accel.

Bird

Allegro \( \frac{d}{= \frac{32}{32}} \)

\[
\begin{array}{c}
\begin{array}{c}
D \quad A7 \\
D \quad D
\end{array}
\end{array}
\]

\[
\begin{array}{c}
\begin{array}{c}
A7 \\
D
\end{array}
\end{array}
\]

accel.
Hands

Adagio

D A7 D A7 D

p

D A7 D A7 G A7 D D A7 D A7

mp

D A7 frog
D A7 D A7 D D

mf

Fish

Andante

A

p

A

E7 A
Elephant

Andante \( \text{\#} = 90 \)

Rabbit

Andante \( \text{\#} = 92 \)
Appendix IX
Song Picture Symbols

Set 1
Set 2

- עכבש
- דבורה
- כוכב
- ארוג
- צפרדע
- עץ
Set 3

פרפר
אפרוח
אנז
פיל
רכבת
דג