A retrospective practice based evaluation of music therapy: A single-case study of a four-year-old girl with Rett syndrome—Rebecca’s story

Simon Hacketta,⁎, Cindy-Jo Morisona,⁎, Carol Pullenc

a Arts Therapies Team, Northumberland, Tyne and Wear NHS Foundation Trust, Newcastle upon Tyne, UK
b Music Therapy Advisor to Rett, UK
c Parent, Alnwick, UK

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ABSTRACT

Introduction: Previous research has found that music therapy (MT) with people who have Rett syndrome (RS) benefits physical, communicative and social functioning. Individual MT has been shown to provide conditions which can stimulate intentional communication and increase opportunities for social interaction.

Method and results: Retrospective video analysis was used to assess in-therapy change in hand function and turn-taking during the first six months of MT with a four-year-old child with RS. Trend analysis using Statistical Process Control charts showed an improvement in the frequency of her hand use and turn-taking in MT.

Conclusion: The paper discusses the techniques used in MT that provided opportunities for the child with RS to increase non-musical skills related to functional hand use and social interaction. Methods for improving the practice-based evaluation of MT in rare disorders such as RS are also presented.

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Music therapy

Rett syndrome (RS) is a neurodevelopmental disorder characterised by mental impairment, loss of expressive speech, deceleration of head growth (microcephaly) and loss of acquired skills (Huppmke, Laccone, Kramer, Engel, & Hanefeld, 2000). RS affects girls almost exclusively, estimates of its prevalence is around 1 in 10,000–15,000 girls (Hagberg, 1983; Zhang & Minassian, 2008). It is associated with mutations in the MECP2 gene on the X chromosome (Amir, Van den Veyver, Wan, Tran, Francke, & Zoghbi, 1999; Huppmke, Bohlander, Kramer, Laccone, & Hanefeld, 2002). The process of X chromosome inactivation and the particular MECP2 mutation are felt to contribute to the variation in the severity of the clinical features of RS. Developmental regression follows a period of apparently normal development with loss of purposeful hand movements and associated with hand stereotypies displayed as hand wringing, tapping, licking, sucking and complex finger patterns (Fitzgerald, Jancovic, & Percy, 1990). This can occur from about eight months to four years when children with RS gradually lose hand function (Lotan & Ben-Zeev, 2006).

Hand stereotypies in individuals with RS are considered to have a role in the complex process of loss of hand use (Temudo et al., 2007). Despite observation that voluntary hand use can be absent or very poor in children with RS, it is of interest that some spontaneous useful movements have been observed to occur in response to external stimuli, for example, movements to music (Kerr, 2008).

Motivation and social interaction have been seen to improve in individuals with RS in response to music therapy (MT). Positive responses to individual MT have been reported in work with 30 girls with RS treated at the Neurological Hospital of Vienna, this included work with a three-year-old girl with RS in 1972 (Wesecky, 1986). The regularity of this therapy and the sensitivity of the therapist in treating each girl individually were identified as important features for the effects to work in improving motivation and function. In a review of the literature music therapy was found to have evidence of stimulating many aspects of development in people with RS including choice making, enhancing vocalisation, function and hand usage, improved eye contact, communication and emotional expression (Lotan & Ben-Zeev, 2006). Individual MT is regarded by some clinicians and researchers as an essential part of communication assessment and therapy for people with RS, supporting learning and development (Elefant, 2005; Kerr, 2008; Sigafoos et al., 2009). Music has also been shown to have a measurable effect on brainstem autonomic functions in people with RS (Bergstrom-Isacsson, Juli, & Witt-Engerstrom, 2007).

⁎ Corresponding author at: The Arts Therapies Team, Psychological Services, Northumberland, Tyne and Wear NHS Foundation Trust, Northgate Hospital, Morpeth NE61 3BP, UK. Tel.: +44 01670 394886.
E-mail address: simon.hackett@ntw.nhs.uk (S. Hackett).

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A systematic review of communication assessment for individuals with RS (Sigafos et al., 2011) identified eight common themes reported to have communicative functions, including (a) eye pointing/eye gaze, (b) body movements, (c) leading, (d) clapping, (e) reaching, (f) pushing away items, (g) vocalisation, and (h) tantrums. The investigation of individual communicative functions of children with RS continues to be an important area of study. The use of traditional single-case designs are sometimes necessary in neuropsychological rehabilitation when patients present with rare syndromes that limit group based studies (Wilson, 1987).

In the present study a retrospective video analysis of music therapy with a four-year-old girl with RS was carried out to assess if hand function and turn-taking were influenced during the first six months of treatment.

### Single-case design

Whilst single-case designs are a tried and tested research approach (Kazdin, 2011), traditional single-case withdrawal or reversal (A–B–A–B) designs are often abandoned in clinical practice on practical, ethical, and humanitarian grounds, for example, it is not preferable to withdraw a beneficial treatment (Callahan & Barisa, 2005). The constraints of clinical practice can also limit the adequate measurement of a baseline condition prior to the introduction of therapy. In this study we have attempted to use an alternative single-case approach which can be applied in the assessment of MT outcomes for individuals with RS in routine clinical settings. The use of retrospective video-analysis of behaviour-observed in-treatment, statistical analysis of data, and parental testimony have been corroborated using a combined approach.

The use of the combined approach to retrospective evaluation draws from a number of sources and utilises video-analysis observations that are then subjected to statistical analysis. There are examples of studies of RS child development that have used video-analysis successfully (e.g., Downs et al., 2008; Fyfe et al., 2007). Observations of turn-taking and hand function are also found in MT studies, such as (Holck, 2004; Wigram & Lawrence, 2005). Statistical process control has been recommended as a scientifically validated method which can help identify significant events and treatment effects in clinical practice (Callahan & Barisa, 2005). We have also drawn on the value of parental testimony in assessing functional progression in RS. Parental testimony has been included in MT case studies, for example (Beathard & Krout, 2008). The addition of parental testimony, combined with the previously mentioned methods, has the potential to add a meaningful account of an individual's progress or otherwise, that can sometimes be missed by formal tests. It has been noted that:

Classical developmental tests fail to register the small meaningful advance which parents report, in particular the changes in the nature of stimuli relevant to the girls, the way the girls express that they have learned something and their content (world of interest) of what they have learned (Demeter, 2000, p. 230).

The study aimed to use empirical trend analysis to assess behavioural observation data from retrospective MT videos of Rebecca, a child with RS. Trend in functional hand use and social interaction, indicated by observations of turn taking, were then corroborated with a parental testimony (Morison, Pullen, Cardwell, & Hackett, 2008).

### Subject

#### Case report

Rebecca was a four-year-old girl with a clinical diagnosis of RS which was confirmed on genetic testing demonstrating a deletion in exon 4 of the MECP2 gene (c.1101-1201 del. 101). Rebecca’s mother has given a descriptive account of events preceding diagnosis:

By the age of twelve months, her pincer grip and eyesight was so good that you couldn’t leave a piece of fluff on the floor without Rebecca being able to spot it and pick it up. In fact she was notorious for this. At this stage in her life, Rebecca was a happy and contented child who could keep herself entertained as well as happily interacting with others around her. She was very vocal and babbled lots and could easily transfer objects from hand to hand. Over the next few months, she took great delight in watching and responding to children’s DVDs, could hold drumsticks to make lots of noise and could even hold a book in both hands and look at the pictures. In fact, she was just like any other children of her age. Almost overnight, Rebecca completely lost her purposeful hand movements. She couldn’t use her drumsticks anymore. Bottles of juice were cast aside after a quick drink. Some people, I think interpreted this as behaviour as that of a naughty child. Rebecca turned from being a happy, contented delightful little child into an insular, miserable one. It seemed like an eternity for us but for six months, Rebecca refused to give us any eye contact. She appeared to prefer her own company. This was a distressing period for all. At this time, Rebecca’s hand movements started to manifest (Morison et al., 2008).

Rebecca was referred for MT at the age of four years old by her paediatrician at the request of her parents. Individual MT sessions lasting up to 30 min were provided by a qualified UK health and care professions council registered Music Therapist and routinely videoed. Rebecca’s mother was able to observe early MT sessions in the treatment room from behind a one-way mirror. Each MT session included a familiar hello and goodbye song (Elefant & Wigram, 2005) and a period of improvisation based upon the Alvin model of free improvisation therapy (Bruscia, 1987). During this period of free improvisation the process “is strictly musical in that all of the client’s therapeutic work centres around listening to or making music. Verbal discussion and other art forms are used but they are secondary to musical activity” (Bruscia, 1987, p. 76). Rebecca had the freedom within this period of free improvisation to create her own beat, rhythm and dynamic. The therapist’s involvement during these improvisations was led by Rebecca through eye contact or touch. When improvising, the therapist had a supporting role, following and not dictating the next “musical move”. Use of this model provided opportunities for Rebecca to make choices independently and establish an “equal terms” relationship (Bruscia, 1987).

Decreased hand stereotypies, and increased purposeful hand use were encouraged by placing beaters with padded handles, for easier grip, within Rebecca’s view. Repeated songs and action rhymes were used to stimulate Rebecca’s voice output and increase vocalisation. Visual aids, objects, instruments, and verbal/vocal prompts were used by the therapist to encourage choice making and promote turn-taking.

### Methods

A single-case retrospective video-analysis was carried out during the first six months of MT: this encompassed 14 individual treatment sessions. Each session video was reviewed by the therapist for two types of behaviour; (a) when Rebecca clearly picked up an instrument, the frequency of observations of “holding”, and (b) “turn taking” included observations of Rebecca vocalising, eye-pointing or chest-patting within a distinct sequence of interaction with the therapist. Video analysis was then checked for inter-rater reliability.

The frequency of observations in each session was counted and then plotted on statistical process control (SPC) charts to assess
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