

Case Report

Efficacy of hand arm bimanual intensive therapy on upper limb co-ordination in a child with Rett syndrome: a case report

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ABSTRACT

Rett syndrome (RTT) is a severe, progressive, neurodevelopmental disorder, which affects predominantly females. RTT is characterized by developmental regression of spoken language and hand use that, with hand stereotypies and impaired ambulation, constitute the four core diagnostic features. Children with RTT exhibit significant impairments in purposeful hand use and bimanual coordination due to neurodevelopmental regression, yet structured bimanual intervention protocols are sparsely reported. The aim of the study is to find out the efficacy of hand arm bimanual intensive therapy on upper limb co-ordination in a child with RTT. This study involved a child of age 4 years diagnosed with RTT, selected based on predefined inclusion and exclusion criteria. Prior informed consent was obtained from the parent/caregiver of the subject. A pre-test assessment was conducted to evaluate upper limb coordination by bimanual fine motor function classification. The child underwent hand arm bimanual intensive therapy for 4 weeks, with session conducted of 35-40 minutes per day, 4 sessions per week. Following the intervention period, the post-test was assessed by same outcome measure as in pre-test. Following the four weeks of intervention, the participant showed progressive improvement in upper limb coordination and bimanual functional performance, with observable gains in voluntary use of hand. The magnitude of improvement ranged from mild to moderate as suggested in bimanual fine motor classification, suggesting positive effects of hand arm bimanual intensive therapy. Hand arm bimanual intensive therapy improved upper limb coordination in child with Rett syndrome following a 4-week intervention.

Keywords: Hand arm bimanual intensive training, Neurodevelopmental disorder, Rett syndrome, Upper extremity

INTRODUCTION

Rett syndrome (RTT), a rare and progressive neurodevelopmental disorder primarily impacting females, has evolved from its first discovery as a distinct clinical entity. As the primary cause of RTT was identified to be related to mutations within the MECP2 gene, this gene was shown to express the protein MeCP2. These proteins are abundant in the brain and act to regulate gene expression. Since being described clinically as a specific disease entity, the diagnostic criteria have undergone numerous revisions. Approximately one in ten thousand live female births will develop RTT. RTT is most commonly due to pathogenic variants in the MECP2 gene. This gene expresses the MeCP2 protein, which is highly expressed in the brain to regulate gene expression.¹

Clinically, RTT progresses over time. A child develops normally during the first six months of life. After that, the child undergoes a regression characterized by stunted head growth, loss of ability to speak, abnormal breathing patterns, reduced mobility and replaced intended hand function with repetitive hand movements. Children with conditions like RTT may exhibit bilateral coordination disorders and/or other impairment in addition to their unilateral deficit.

Hand-arm bimanual intensive therapy (HABIT) was developed as a method of improving coordination between both hands. It uses intensive practice as part of carotid intima-media thickness (CIMT); however, the application is through task-based bimanual play or functional activities. HABIT is based on the theories of motor

learning including practice-specificity and feedback and neuroplasticity via repetition, increased movement complexity and motivation.²

Children with condition such as RTT often experience significant impairments in bimanual coordination that extend beyond their unilateral deficits. To address these challenges, HABIT has been developed, grounded in the principles of motor learning and neuroplasticity.³

HABIT is a form of practical bimanual training to develop coordinated use of each arm. Each task will require extensive practice of CIMT constrained induced movement therapy but will involve additional components of bimanual play and functional actions. The program was developed on principles of motor learning, including practice specificity and feedback, and principles of neuroplasticity, including repeated movement, increased complexity of movement, and motivation to elicit movement-induced changes in the brain.⁴

Children with RTT exhibit significant impairments in purposeful hand use and bimanual coordination due to neurodevelopmental regression, yet structured bimanual intervention protocols are sparsely reported. There is a lack of comparative, case-based clinical studies evaluating the feasibility and functional outcomes of a uniform HABIT protocol when applied to a paediatric condition of neurological origin. Establishing evidence for HABIT in such population is essential to guide clinical decision-making and broaden its therapeutic application.⁵

Therefore, the present study aims to address this gap by investigating the effects of HABIT on functional upper limb use in children with RTT thereby contributing to evidence-based paediatric physiotherapy practice.

CASE REPORT

The purpose of this case report was to evaluate the effectiveness of HABIT on upper limb coordination in a child diagnosed with RTT. A child who attended the physiotherapy outpatient department and was diagnosed with RTT through clinical and genetic evaluation was selected for the study. The subject was recruited based on the inclusion and exclusion criteria. Prior informed consent was obtained from the parent/caregiver before the

commencement of the study. Materials used for the assessment included the bimanual fine motor function (BFMF) classification system.

A baseline assessment of upper limb coordination and bimanual hand function was carried out using the BFMF classification. Following the pre-intervention assessment, the child underwent HABIT for a period of four weeks, with four sessions per week and each session lasting approximately 35-40 minutes. The intervention consisted of sensory and regulatory preparation, guided bimanual engagement activities, functional play tasks, and positive reinforcement techniques aimed at improving purposeful hand use and upper limb coordination. Post-intervention assessment was performed at the end of four weeks using the same outcome measure to evaluate changes in bimanual fine motor function and upper limb performance.

Outcome measures

Bimanual fine motor function classification

The BFMF classification is a validated tool used to assess hand function and fine motor abilities in children. It evaluates a child's ability to grasp, hold, manipulate objects, and perform bimanual tasks during daily activities. The BFMF classifies hand function into five levels (I-V) based on the quality, effectiveness, and independence of hand use.

Due to its good reliability and validity, the BFMF is widely used in clinical practice and rehabilitation research to assess functional hand performance and monitor treatment outcomes.⁵

Classification of BFMF levels

Level I

Handles objects easily and performs fine motor tasks efficiently with both hands.

Level II

Handles most objects independently but with slightly reduced speed or quality.

Table 1: Techniques and phases in intervention.

Step	Phase	Duration	Activities and techniques
1	Sensory and regulatory prep	10 minutes	Purpose: Decrease stereotypies and prepare hand - deep pressure on shoulders and arms, compressions, and low vestibular inputs (rocking or sitting on lap)
2	Guided bimanual engagement	10 minutes	Purpose: Active use both hands - position the child so both hands are accessible, guide movements without forcing, encourage simultaneous or complementary hand use (e.g.: holding a toy with both hands, pulling apart velcro toys)
3	Functional play/activity	10 minutes	Purpose: Meaningful, visually motivating tasks, example: holding a box with one hand while opening lid with another, and focus on activities the child finds rewarding

Level III

Requires assistance or adaptations for more complex fine motor activities.

Level IV

Limited ability to manipulate objects and requires considerable support.

Level V

Severe limitations in hand function with minimal ability to perform purposeful object manipulation.

Intervention

Techniques and phases in intervention is shown in Table 1.



Figure 1: Giving HABIT to child with RTT with a therapy ball.

RESULTS

The results indicate an improvement in bimanual fine motor function following the intervention. Based on the BFMF score, improved from level V to level IV, showing better hand function and coordination. The mean values also reflect a positive change in the case, indicating enhanced ability to perform bimanual activities. Overall, these findings suggest that the intervention was effective in improving fine motor skills and functional hand use in the patient.

DISCUSSION

The present case report was conducted to evaluate the effectiveness of HABIT on upper limb coordination in a child diagnosed with RTT. Following four weeks of intervention, improvements were observed in purposeful hand use, bimanual coordination, and overall functional performance as assessed using the BFMF classification.

These findings suggest that HABIT may be beneficial in enhancing upper limb function in children with RTT.

RTT is a progressive neurodevelopmental disorder characterized by regression of acquired motor and communication skills, loss of purposeful hand use, and the development of stereotypic hand movements. In the present case, the child demonstrated impaired functional hand use and difficulty performing bimanual activities. Following HABIT, an improvement in voluntary hand movements and engagement in functional tasks was observed. These findings are consistent with those reported by Gold et al who highlighted the importance of therapeutic interventions targeting functional hand use and participation in children with RTT.⁷

The improvement observed in this study may be attributed to the principles of motor learning and neuroplasticity on which HABIT is based. HABIT emphasizes intensive, repetitive, task-specific, and goal-directed bimanual activities that encourage active participation and functional use of both upper limbs. Similarly, Gordon et al reported significant improvements in bimanual coordination and functional performance following HABIT interventions in children with upper limb impairments.²

Furthermore, Smeets et al described RTT as a condition associated with significant motor dysfunction and reduced hand function, which negatively impacts activities of daily living.

The present findings indicate that structured bimanual training may facilitate improved motor performance and encourage purposeful use of the upper limbs despite the neurological limitations associated with the disorder.⁸

The BFMF classification was used as the outcome measure in this study. Post-intervention assessment demonstrated improvement in fine motor abilities and functional hand use, suggesting that BFMF is a useful tool for documenting functional changes in children with motor impairments.⁵

Limitations

Despite the positive outcomes observed, this case report has certain limitations. The study included only one participant and the intervention period was limited to four weeks. Long-term follow-up was not conducted to determine the retention of treatment effects. Further studies with larger sample sizes and extended intervention durations are recommended to establish stronger evidence regarding the effectiveness of HABIT in children with RTT.

CONCLUSION

Overall, the findings of the present case report suggest that HABIT may be an effective intervention for improving

upper limb coordination and functional hand use in children with RTT.

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