Effect of Hand Splints on Stereotypic Hand Behavior of Three Girls with Rett Syndrome

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The purpose of this multiple baseline study was to examine the effect of bilateral hand splints on the persistent stereotypic hand movements of three adolescent girls with Rett syndrome. Among the most characteristic features of Rett syndrome are stereotypic hand-wringing and hand-biting behavior and loss of previously acquired functional hand skills. The hand splints used in this study consisted of cuffs encircling the palm that positioned the subjects' thumbs in abduction. Duration percentages of subjects' stereotypic hand behavior and functional hand use were calculated from five-minute videotaped segments recorded during a finger-feeding condition and a free-time condition. All three subjects demonstrated a decrease in the amount of time spent in stereotypic hand behavior after application of hand splints, and one subject showed an increase in finger-feeding skills while wearing hand splints. Limitations of the study are discussed, and suggestions for clinical application and future research are offered.

Key Words: Hand; Medical conditions; Orthotics/splints/casts, upper extremity; Stereotyped behavior.
conditioning requires the presence of a "behavior manager" with the child to provide intervention. Iwata et al did not indicate whether the reduction in hand-biting behavior could be generalized to situations where the child was not monitored by a manager. The use of hand splints, should they prove effective, would be advantageous because the child could experience the benefits of intervention independent of a manager. In addition to cost effectiveness, the use of hand splints has the potential advantage of allowing classroom staff to spend more time in instructional programs rather than in response-interruption programs.

Hand splints have been reported clinically to decrease the frequency of stereotypic hand behaviors in girls with Rett syndrome, but no systematic examination of this treatment strategy has yet been completed. Hanks commented that splinting is found to be successful in interrupting hand-to-mouth, and to a lesser degree, hand-wringing [behavior], thus allowing the girls to direct their attention outward to notice toys and people rather than having their attention totally dominated by their hand wringing.

Because hand splints could potentially reduce stereotypic hand behavior and influence the ability of girls with Rett syndrome to interact with their environment, this multiple baseline study explored the effect of hand splints on the stereotypic hand behavior of three girls with Rett syndrome.

An additional purpose of this study, based on the findings of Iwata et al and others, was to examine whether an increase in the subjects' purposeful hand usage would coincide with or follow the reduction of stereotypic hand behavior. As Hanks stated, "Indeed, observation makes it fairly evident that the girls would like to do something else with their hands but cannot break through the hand wringing to do so."

METHOD

Subjects

The three subjects were enrolled five days a week in a special education program for students with severe or profound handicaps at a university-affiliated campus school. The subjects attended school 5.5 hours a day, and in addition to their regular classroom program, they were treated by a communication disorders specialist, a physical therapist, and an occupational therapist. Subject B attended school 4.5 hours a day during the first half of the study because of special transportation needs. Written informed consent was obtained from a parent of each subject.

All three subjects were diagnosed with Rett syndrome at the Child Development and Mental Retardation Center. Subject C was diagnosed with Rett syndrome in 1983 at the age of 13 years, one of the earliest cases of Rett syndrome diagnosed in the United States. Subjects A and C were diagnosed with Rett syndrome in 1984 at 11 and 14 years of age, respectively.

Subject A was a 13-year-old girl whose stereotypic hand behaviors began at 15 months of age. Before this study began, her hand behaviors consisted of bilateral "pill-rolling" motions of her fingers against her thumb and palm and gentle clapping motions of her fists. Subject A had worn a resting hand splint occasionally to maintain left wrist extension, but because her wrist range of motion was measured within normal limits, use of this splint was discontinued during this study. Subject A's previous levels of functional hand use were very limited. She could hold a straw to her mouth after it had been placed in her hand, and she was learning to reach with her right hand to grasp a small piece of food held in front of her at eye level. Subject A did not use these functional skills consistently.

Subject B was a 16-year-old girl who began showing stereotypic hand behavior at about 2 years of age. At the start of this study, her hand movements consisted of slow repetitive hand-wringing, hand-clapping with flexed fingers, and hand-mouthing behaviors. Subject B had never worn an upper extremity orthosis for reduction of stereotypic hand behavior or any other reason. Before intervention, Subject B could finger feed herself independently but was laboriously slow (eg, she took about one hour to eat a sandwich and piece of fruit). She could also activate simple switches by hand.

Subject C was also 16 years old and began showing stereotypic hand movements at about 1 year of age. Before this study began, her hand movements consisted of repetitive hand wringing with her hands together, bilateral hand squeezing with her hands apart, and hand-mouthing behaviors. Subject C had previously been fitted with a unilateral hand splint, similar in design to the splints used in this study, but its use was discontinued three months before this study began because the splint was ineffective in reducing stereotypic hand movements. Bilateral splinting may have been necessary to reduce the subject's stereotypic hand movements. Subject C's previous levels of functional hand use were limited to grasping the handles of a wheeled walker and occasionally reaching to grasp an object held in front of her at eye level.

Materials

Three pairs of thumb abduction splints were fabricated by an occupational therapist experienced in splinting techniques. The splints were constructed of Polyform® splinting material. This material was used because it is thin, smooth, and rigid but very moldable when heated. Because the splints were fitted to accommodate the contours of three different pairs of hands, they were slight variations of a basic design. The splints resembled a cuff circling each palm and positioned the thumb out of the palm into abduction (Fig. 1). The splints restricted excessive opposition of the thumb across the palm but did not functionally restrict the subject's grasp or movements of the fingers and wrist. Wearing time was gradually increased until the subjects wore the splints every school day from 9 AM to 2:30 PM.

Response Definitions

Stereotypic hand behavior was defined as 1) hands in contact with each other and moving in any way (washing, wringing, or clapping movements), 2) hands not in contact but one or both hands engaged in a repetitive squeezing or pill-rolling motion, or 3) one or both hands in contact with the subject's lips or tongue unless the subject was putting a piece of food into her mouth. One hand could be resting quietly as long as the other hand fit one of the above criteria. Behavior ceased to be considered stereotypic if both hands discontinued the movements described above or if both hands were motionless for more than two seconds.

Functional hand use was defined as the subject's hand contacting a piece of food and moving toward the subject's mouth. The definition excluded time when the subject held...
food but ceased to bring it toward her mouth for more than two seconds. As long as the subject had one hand in the process of grasping a piece of food, bringing it to her mouth, or placing it into her mouth, functional hand use would be scored, even if the subject's other hand was engaged in stereotypic hand movement.

The definitions for stereotypic hand behavior and functional hand use were exclusive. Both conditions could not occur at the same time. In addition, periods of time could exist when the subjects’ behavior would not fit either of the two conditions (eg, when the subject’s hand had left her mouth but had not yet resumed stereotypic movements or picked up another piece of food).

**Experimental Design and Phases**

This study used a multiple-baseline, across-subjects design. During baseline phases, subjects did not wear hand splints and could initiate functional hand use or perform stereotypic movements as they preferred while in the classroom. During intervention phases, subjects wore a splint on both hands for increasingly longer periods of time. As during the baseline phases, hand behaviors (stereotypic or functional) during intervention phases were not interrupted nor was any attempt made to reinforce them. A brief withdrawal phase (return to baseline condition) was conducted for all subjects at the completion of the study to identify any maintenance effects of the hand splints. Subjects did not wear their hand splints during the withdrawal phase and could perform whatever hand behavior they preferred.

In keeping with multiple-baseline design requirements, hand splints were applied in a sequential fashion to each subject only after stability on a behavioral level or a trend was established. After 15 days of the baseline condition, Subject A began wearing her splints. Baseline data continued to be collected for the other two subjects. Wearing time for Subject A was gradually increased over the next nine days. The rate of increase for all subjects was determined by skin tolerance. The appearance of red marks that did not fade within 15 minutes after splint removal resulted in smaller increments of increased wearing time. On Day 29 of the study, the hand splints were introduced to Subject B. Wearing time for Subject B was increased over the next four days. Baseline conditions continued for Subject C, and on Day 43 she began wearing hand splints. Wearing time for Subject C was gradually increased over the next seven days. Fifty-four days after the start of the study, all three subjects were wearing hand splints the entire time they were at school. The subjects' spring vacation coincided with Days 59 to 73, and during this period, none of the subjects wore their splints. This withdrawal phase continued during their first week back at school (Days 74–78). The subjects began wearing their splints again during the second week after spring vacation. The withdrawal phase for Subject B began before spring vacation on Day 55. Her withdrawal phase began early so that we could obtain enough data to establish internal validity for functional hand use.

**Data Collection**

Videotapes were recorded during the afternoon of each school day with each subject seated at a table in a quiet part of the classroom with minimal visual distractions (e.g., few individuals moving about within the subject’s visual field). A dish containing pieces of a cracker was placed on the table directly in front of the subject within easy reach. The subject was positioned with both upper extremities placed on the table so that both hands were visible to the camera at all times. No instructions or prompts were provided so that subjects would independently initiate and maintain any hand behavior, whether stereotypic or functional. Each videotaping session lasted five minutes. The use of a video recorder enabled durations of stereotypic and functional behavior to be timed separately with a digital stopwatch and thus eliminated the
need for the investigator to simultaneously measure two behaviors during direct observation.

Duration of stereotypic movements also was recorded during a five-minute free-time period just before the finger-feeding condition where the subject was seated in a chair listening to music through headphones. Duration measurements were taken from videotapes recorded three days a week (Tuesday, Thursday, and Friday). The free-time condition provided an opportunity to examine any differential effects of the hand splints specific to the demands of the situation for production of functional hand use. The furniture, materials, and positioning of subjects and equipment were kept constant throughout all of the data collection sessions and the intervention phase. Subjects wore their hand splints during each data collection session.

Reliability

Interobserver agreement. Interobserver agreement was assessed every fifth day of data collection with one of the authors (G.M.N.) as the primary observer and the subjects' occupational therapist as the second observer. The observers simultaneously viewed each videotaped segment but were positioned in the room so that they could neither see nor hear how the other observer was scoring the videotaped segment. Neither observer was blind to the purpose of the study. Percentages of interrater agreement were calculated for all duration conditions (stereotypic movement during presentation of food, functional hand use, and stereotypic movement during free time) for all three subjects using the following formula:

\[ \text{Percentage of Interrater Agreement} = \frac{S_o}{S_o + S_i} \times 100 \]

where \( S_o \) is the smaller of two observers' scores and \( S_i \) is the larger of the two observer's scores. Percentages of interrater agreement for stereotypic movement during presentation of food ranged from 67% to 100% (\( \bar{X} = 92.5\% \)). Scores fell below 80% on 2 of 27 occasions (7.4%). Percentages of interrater agreement for functional hand use ranged from 90% to 99% (\( \bar{X} = 95.6\% \)). Percentages of interrater agreement for stereotypic movement during free time ranged from 64% to 99% (\( \bar{X} = 91\% \)). Scores fell below 80% on 4 of 27 occasions (14.8%).

Point-by-point agreement was also calculated every five data collection days using the same videotaped sessions that were used to calculate percentages of interrater agreement. This method of assessing interrater reliability was used in addition to percentages of interrater agreement because percentages of interrater agreement reflect only agreement on the total amount of behavior observed, not whether the observers agreed on any particular instance of performance. Each 5-minute session was divided into 10-second intervals, and each observer scored whether stereotypic behavior or functional hand use, or neither, had occurred at the end of each interval. Point-by-point agreement was calculated by dividing the number of agreements by the number of intervals and multiplying the quotient by 100. Point-by-point reliability scores ranged from 84% to 100% (\( \bar{X} = 93.9\% \))

Procedural reliability. Procedural reliability was assessed during all phases of the investigation. This process was used to examine the reliability with which experimental procedures were followed. The classroom teacher assessed the experimental environment weekly to determine whether it was set up in a consistent manner. The data collector was unaware of monitoring occasions. A check list of setup requirements was provided for the classroom teacher to determine consistency (eg, proper placement of the video camera in relation to the subject, both of the subject's upper extremities visible to the camera, subject listening to music through audio headphones during the free-time condition). The experimental environment was reproduced correctly on all monitoring occasions. Wearing time of the hand splints for all subjects was also monitored to ensure appropriate use. Actual wearing time was monitored daily and was consistent with the established wearing schedule on 93%, 95%, and 92% of the intervention days for Subjects, A, B, and C, respectively. Actual wearing time on days that were inconsistent with the schedule deviated by an average of 1.6 hours.

RESULTS

Stereotypic Hand Behavior

The data for stereotypic hand behavior of the three subjects during the feeding condition are shown in Figure 2. The percentage of time Subject A spent performing stereotypic hand movements during baseline sessions remained fairly stable from day to day (\( \bar{X} = 69.8\% \)). The hand splints were introduced on Day 16, and during the intervention phase, Subject A demonstrated a slow, gradual decrease in stereotypic hand behavior. During the last five days of the intervention phase, her mean level of stereotypic hand behavior had decreased to 37.2%. The mean duration of stereotypic hand behavior for the entire intervention phase was 45.8%. During the second baseline (withdrawal) phase that followed the 15-day spring vacation, Subject A's stereotypic behavior returned to a level higher than the original baseline level (\( \bar{X} = 79\% \)). Three data collection sessions had been planned for this phase, but data on one of those sessions was not obtained because of Subject A's unusually extreme sleepiness.

The stereotypic behavior of Subject B during the baseline phase was fairly stable with no change in trend when hand splints were introduced for Subject A. The mean duration of stereotypic hand behavior for Subject B during the baseline phase was 54.9%. After application of the hand splints on Day 29, she displayed an immediate and marked decrease in stereotypies (\( \bar{X} = 6.2\% \)). During the baseline phase, Subject B would frequently resume stereotypic hand movements between each bite of food. During the intervention phase, she only occasionally engaged in stereotypic hand movements between bites, and her hands tended to rest quietly, or she attempted to pick up another piece of food. During the second baseline phase, her stereotypic hand behavior immediately increased to a mean level beyond that achieved during the original baseline phase (based on data points both before and after spring vacation; \( \bar{X} = 69.7\% \)).

The baseline data for Subject C were quite variable but did not display any particular trend and did not appear to be affected by the introduction of hand splints to Subject A or Subject B. Subject C's mean duration of stereotypic hand behavior during the baseline phase was 62.2%. After application of her hand splints on Day 43, Subject C also displayed an immediate decrease in hand movements with a mean duration of 13.7% during the intervention phase. Despite the considerable variability in Subject C's baseline phase data, only two of the baseline data points overlapped into the range of the intervention phase data. During the withdrawal phase after the spring vacation, Subject C's stereotypic hand behav-
behavior returned to near-baseline levels ($\bar{X} = 44.3\%$). No overlap of data points existed between the intervention phase and the second baseline phase.

Figure 3 illustrates the stereotypic hand behavior of the three subjects during the free-time condition. Subject A demonstrated an upward trend in stereotypic hand behavior during the baseline phase ($\bar{X} = 75.8\%$). As in the feeding condition, she responded to the application of her hand splints with a slow, gradual decrease in stereotypic hand behavior. During the last five days of the intervention phase, her mean level of stereotypic hand behavior had decreased to 30%. The mean duration of stereotypic hand movements during the entire intervention phase was 48.1%. Levels of stereotypic hand behavior during the second baseline phase exceeded the original baseline level ($\bar{X} = 84\%$). The hand splints appeared to have no substantial differential effects dependent on the conditions of the environment (feeding or free time) for Subject A. The levels of stereotypic hand behavior emitted through all phases of the free-time condition were slightly higher than the levels in corresponding phases of the feeding condition.

Subject B showed very stable day-to-day behavior during the baseline phase ($\bar{X} = 77.4\%$). She displayed an immediate
Fig. 3. Subjects' stereotypic hand movements during free time. (Duration = percentage of time response occurred during data collection interval.)

decrease in stereotypic hand behavior after introduction of hand splints during the intervention phase ($\bar{X} = 20.4\%$). The withdrawal phase brought an increase in stereotypic behaviors higher than the original baseline level ($\bar{X} = 87.2\%$). As with Subject A, the only noticeable difference between the feeding and free-time conditions for Subject B appeared to be slightly elevated levels of stereotypic hand behavior during all phases of the free-time condition.

The stereotypic hand behavior of Subject C was quite variable during the baseline phase of the free-time condition. No apparent trend or change in the data, however, occurred with introduction of hand splints to Subjects A and B. The mean duration of stereotypic hand behavior during the baseline phase was $68.4\%$. During the intervention phase, Subject C exhibited an immediate decrease in stereotypic hand movement ($\bar{X} = 18.4\%$). Despite the variability of the data in the baseline phase, data points did not overlap between the baseline and intervention phases. The withdrawal phase demonstrated an increase in the level of stereotypic hand behavior toward the original baseline level ($\bar{X} = 37\%$). One data point from the second baseline phase overlapped the range of data points from the previous phase. As with the other subjects, no difference in hand splint effectiveness existed between the feeding and free-time conditions. Subject C did display slightly
elevated levels of stereotypic hand behavior during the first baseline and intervention phases of the free-time condition.

**Functional Hand Use**

Before introduction of the hand splints, Subject A and Subject C did not use finger-feeding skills. Subject B could finger feed herself independently but very slowly. Subjects A and C did not initiate independent finger feeding after application of the splints, although their hand behavior during intervention phases was no longer dominated by stereotypic hand movements. Subject B, however, did demonstrate an immediate increase in finger feeding after introduction of the hand splints. Figure 4 contains the data for the functional hand use of Subject B. The mean duration of finger feeding of Subject B during the baseline phase was 31.8%. The occurrence of finger feeding increased dramatically after introduction of the splints on Day 29. During the intervention phase, the mean duration of functional hand use increased to 66.6%. The amount of time Subject B required to eat a sandwich and piece of fruit decreased from about 60 minutes to about 45 minutes. The extreme data point corresponding to Day 51 was very atypical of Subject B’s behavior. She appeared to be ill and ate nothing the entire school day. After removal of the hand splints, the duration of finger feeding in Subject B during the second baseline phase returned almost to her original baseline level ($\bar{X} = 23.3\%$).

**DISCUSSION**

A decrease in stereotypic hand behavior occurred in all three subjects after the application of hand splints; for two of the subjects, the effect was both immediate and substantial. This repeated demonstration of the target behavior changing in response to (and not before) application of the intervention makes the influence of extraneous variables implausible. In addition to demonstrating an absence of any lasting effect after removal of the hand splints, the withdrawal phase conducted with each subject provided a second baseline condition. Data obtained in that baseline condition strengthened the internal validity of the investigation, because the stereotypic hand behavior of each subject and the functional hand use of Subject B returned to levels near those achieved during the original baseline phase.

The results of this study were consistent with the findings of Iwata et al in successfully reducing stereotypic hand behaviors in girls with Rett syndrome. Although Iwata et al isolated hand-biting behavior as the target of intervention and our study examined combinations of hand-biting and hand-wringing behaviors, both studies demonstrated that these behaviors can be reduced substantially.

In addition to significantly reducing stereotypic hand behavior, the hand splints were effective in improving the finger-feeding skills (functional hand use) of Subject B. She engaged in finger feeding more frequently and required less time to feed herself after application of the splints. After splint application, Subject B’s finger feeding was no longer interrupted by constant stereotypic hand movements. The stereotypic hand behaviors had possibly been functioning as a competing behavior to purposeful hand use. This result is consistent with the findings of Iwata et al and Bierly and Billingsley that showed an increase in appropriate behavior concurrent with reductions in stereotypic hand behavior.

Hand splints, however, did not produce finger feeding in the two subjects who did not possess finger-feeding skills before this study. Subjective observations by the classroom teacher and the occupational therapist indicated that Subject A’s ability to grasp a piece of food held in front of her at eye level and bring it to her mouth and her ability to grasp a spoon, scoop food onto it, and bring it to her mouth did improve after application of the splints. Because these skills were part of a classroom program, whether instruction or the hand splints caused the reported behavior change is unclear. By decreasing the amount of time the subject spent in stereotypic hand behavior, a state more conducive to learning and practice may have been created. Future investigations should explore whether individuals with Rett syndrome can learn functional hand skills more readily when their stereotypic hand behaviors are reduced with hand splints.

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**Fig. 4.** Functional hand use of Subject B. (Duration = percentage of time response occurred during data collection interval.)
No substantial differential effects of the hand splints specific to the degree of opportunity presented by the environment for production of functional hand use existed. The pattern of response to hand splint application was similar for both the feeding and free-time conditions. In all three subjects, a slight elevation occurred in the levels of stereotypic hand behavior during the free-time condition compared with the feeding condition. The subjects could have spent more time engaged in stereotypic hand behavior when no activity demands were placed on their hands, but this study did not clearly demonstrate that conclusion.

Limitations

One limitation of this study is the possible expectation bias introduced by the primary data collector and the second observer. Neither individual was blind to the purpose of the study. Interrater reliability could be strengthened by having videotaped segments viewed and scored by raters who are blind to the purpose of the study and to which baseline phase the subject was in. The use of such procedures obviously is difficult when the presence or absence of the treatment device (eg, hand splints) is obvious to the viewer. An additional limitation of the study is a possible reduction in the degree to which the results can be generalized to an average classroom environment. We used a quiet part of the classroom that was not typical of the usual activity level of that room. The effectiveness of hand splints may be affected by the amount of distractions in the environment.

Implications for Future Research

The mechanism by which hand splints influence stereotypic hand movements is unknown. Hand movements may create a type of sensory input required by individuals with Rett syndrome, and hand splints may provide that input or alter the need for that input in some way. Another possible explanation was derived from a technique used in the rehabilitation of patients who have experienced an upper motoneuron lesion and display synergistic patterns of muscle spasticity. The position of thumb abduction provided by the hand splints is similar to the reflex-inhibiting posture described by Bobath16 and the thumb maneuver described by Brunstrom.17 The thumb position reverses a component of the total synergy of movement (elbow flexion, wrist flexion, and adduction of the fingers and thumb) and thus creates a “relaxation” of the total synergy. Elongation of shortened muscle groups or joint proprioception, or a combination of both, may inhibit this movement pattern. Future researchers should attempt to identify which characteristic of the splints or process resulting from splint application is responsible for the reduction of stereotypic hand movements.

Future researchers should also investigate whether procedures could be implemented that might allow subjects to discontinue use of hand splints. This study demonstrated no maintenance effects of hand splints over time; however, a gradual reduction in hand splint wearing time, compared with an abrupt termination, could promote a more enduring outcome. Future studies could also assess hand-wringing and hand-mouthing behaviors separately. We noted subjectively that the hand splints used in this study appeared to reduce pill-rolling and hand-wringing behaviors to a greater extent than hand-mouthing and hand-clapping movements. Future studies could assess these differences objectively.

CONCLUSION

The results of this study suggest that the use of hand splints can successfully reduce stereotypic hand behaviors in girls with Rett syndrome. The associated risk of skin breakdown thus can be reduced significantly. Although the functional hand use of one subject improved after application of the splints, children with Rett syndrome should not be expected to begin to spontaneously demonstrate functional behaviors that they were unable to perform before hand splinting. Hand splint application may create a behavior state that is more conducive to learning and practicing functional hand skills. This study was possibly the first to examine the use of hand splints in girls with Rett syndrome, and the study procedures should be replicated to determine generality of results.1 Research based on a single-subject design relies on replication of procedures across settings and subject types to establish the extent to which treatment may be applied successfully to individuals with diverse characteristics in various contexts.

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Postscript. After completion of this study, an initial attempt was made to explore the limits of generality of the hand-splinting procedure by applying hand splints to one additional subject who displayed stereotypic hand movements but did not have Rett syndrome. The subject was a 9-year-old boy with severe mental retardation and tuberous sclerosis. No effect was observed on the subject’s level of hand stereotypic behavior as a function of splinting; however, the subject never tolerated hand splints for more than two hours a day. The data from that investigation are available from the senior author (G.M.N.).

REFERENCES