Promoting object manipulation and reducing tongue protrusion in seven children with Angelman syndrome and developmental disabilities through microswitch-cluster technology: A research extension

Keywords: Angelman syndrome, Microswitch-Cluster Technology, Positive Participation, Social Validation, Quality of Life, Challenging Behavior

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Running Head: AS and MCT

ABSTRACT

We further extended the use of a microswitch-cluster technology to promote object manipulation and to reduce tongue protrusion in seven children with Angelman syndrome. Study I included seven participants with severe to profound developmental disabilities. An ABB^1AB^1 experimental sequence was implemented. During the baselines (i.e., A phases) the technology was available but inactive. During the intervention (i.e., B phase) the adaptive responding was positively reinforced irrespective of the challenging behavior. During the cluster (i.e., B^1 phases) the adaptive responding was contingently reinforced only if it occurred free of the challenging behavior. A long-term follow-up (i.e., 24 months) was conducted. Intervals with indices of positive participation as an outcome measure of the participants' constructive engagement and favorable occupation were additionally recorded. Study II recruited 56 external raters (i.e., equally divided in 4 groups among caregivers, physiotherapists, psychologists, and teachers) in a social validation procedure. Results evidenced the effectiveness and the suitability of the technology to pursue the dual goal (i.e., increasing the adaptive responding and simultaneously decreasing the challenging behavior). All the participants consolidated their learning process and positively participated along the intervention phases. Social raters favorably scored the use of the technology. Educational, clinical, psychological, and rehabilitative implications of the findings were critically discussed. Some useful insights for future research and practice were emphasized.

INTRODUCTION

Angelman syndrome (AS) is a genetic and neurological disorder mainly caused by a deletion in the long arm of the 15th maternal chromosome, first described by the British pediatrician Angelman (1965). Because three of his patients evidenced excessive laughing, jerky movements, severe to profound developmental delays, and flat head disorder associated with physical development abnormalities, they were called "puppet children". Additionally, the E6-AP (i.e., E6 protein encoded by the UBE3A gene) seems to be responsible for the genetic AS (Fisher, Keng, & Ziegler, 2020). The phenotype usually includes ataxia, seizures, tongue protrusion, motor impairments, microcephaly, widely spaced teeth, hyperactivity, prominent chin, restless behaviors, hypo-pigmentation (i.e., blond hairs and light eyes), hypotonia, and wide gait (Heald, Allen, Villa, & Oliver, 2013). Learning difficulties, specific alterations of the electro-encephalogram patterns (EEG), and autism spectrum disorders (ASD) traits are additionally recognizable (Goswami, Sahu, & Singhi, 2018). Furthermore, reduced social interactions, lack of speech and communication disorders, stereotypic movements (e.g., hand flapping) are commonly embedded (Buiting, Williams, & Horsthemke, 2016). Finally, sleep disturbances are frequently observed (Egan, Farrell, Hoey, McGuire, & Lydon, 2020; Trickett et al., 2019).

In light of the above, children with AS may experience a deleterious situation due to their clinical conditions, which may seriously hamper their social image, status, and desirability, with negative outcomes on their quality of life (Hamrick & Tonnsen, 2018). In fact, hyperactivity, disengagement, and anxiety are identifiable among this population. Accordingly, children with AS constantly rely on their caregivers' assistance (Kaskowitz, Dendrinos, Murray, Quint, & Ernst, 2016). To profitably tackle this issue, one may conceive the use of an assistive technology-based intervention (AT; Boot, Owuor, Dinsmore, & Maclachlan, 2018). AT refers to any device, equipment, piece or tool enabling individuals with severe to profound developmental disabilities with independence and self-determination towards their environment (Lancioni & Singh, 2014). Thus, AT minimizes the

existing gap between human capacities and environmental requests (Hoppenbrouwers, Stewart, & Kernot, 2014). Consequently, by using an AT-based program, a person with multiple disabilities will be capable to favorably cope with his/her outside world (Squires, Williams, & Morrison, 2019). A basic form of AT are microswitches (Stasolla, Perilli, & Boccasini, 2016).

Microswitches are electronic sensors capable of capturing a minimal response available in the person's behavioral repertoire and providing brief periods of positive stimulation contingently through a computerized system or an adapted system control unit (Stasolla, Boccasini, et al., 2014). For example, a girl with multiple disabilities laid on her back may be taught to use her hand movements to activate tilt sensors fixed on a grid suspended above her face and receive 7-10 seconds of favorite stimuli contingently during intervention phases (Lancioni et al., 2004). Few basic rules should be observed to ensure the program success. First, a plausible behavioral response already available in the person's repertoire should be identified. Second, an electronic device (i.e., microswitch) capable of detecting the individual's response should be adapted. Third, a highly motivating and rewarding pleasant stimulation to be used as primary and positive reinforcements should be selected. Based on learning principles (i.e., causal association between behavioral responses and environmental consequences), whenever the aforementioned rules are applied, one may reasonably argue that the participant should favorably acquire the functional use of the microswitch to get the independent access to the positive events (Stasolla & Perilli, 2015). Within this framework, microswitch-cluster technology (MCT) is used as a specific rehabilitative intervention focused on pursuing the dual simultaneous goal of promoting an adaptive responding and decreasing a challenging behavior (Lancioni, Singh, O'Reilly, & Sigafoos, 2009). For instance, a boy with cerebral palsy and multiple disabilities may be exposed to a microswitch-cluster program aimed at enhancing arm lifting and reducing dystonic sideway head tilting simultaneously. Both responses were recorded through two tilt devices, fixed on the participant' arm and head respectively, which combined constituted the cluster. Firstly, the participant was exposed to a baseline phase with the technology available but inactive. Secondly, an intervention phase useful to increase the adaptive responding

irrespective of the challenging behavior occurred. Finally, a cluster phase where the adaptive responding was positively reinforced contingently only if it was totally free of the challenging behavior was implemented. Results showed that the participant learned the functional use of the technology (Lancioni, Comes, et al., 2005).

Although the literature on the use of MCT is substantial (Lancioni et al., 2005, 2006, 2008, 2009, 2013; Lancioni, Singh, O'Reilly, Sigafoos, 2011; Lancioni, Singh, O'Reilly, & Oliva, 2014), few studies were targeted on pediatric populations including rare genetic syndromes and/or ASD (Perilli et al., 2019; Stasolla, Perilli, et al., 2014, 2017). To our knowledge, this is the first investigation including an AT-based program focused on children with AS. Thus, by using AS and AT as keywords in SCOPUS database, no records were found. Moreover, studies involving external raters in a social validation assessment (i.e., a standard procedure helpful to corroborate both intervention's clinical and social validities) actually used up to three groups of raters exposed to three experimental conditions. Finally, whenever implemented, a follow-up was extended up to 12 months (Perilli, et al., 2019; Stasolla, Caffò, Perilli, & Albano, 2019 a, b).

As a result, the current investigation, which encompasses two studies, would provide a suitable research extension on this specific topic with the following objectives. Study I represented the first attempt available in the literature to expose seven children with AS and severe to profound developmental and intellectual disabilities to a MCT with the simultaneous dual goal of promoting object manipulation and reducing tongue protrusion. Intervals of indices of positive participation as an outcome of participants' active role, constructive engagement, and quality of life were additionally recorded (Stasolla et al., 2019). A long-term follow up (i.e., 24 months) was carried out. Study II involved 56 external raters, equally divided in 4 groups (i.e., caregivers, physiotherapists, psychologists, and teachers), with at least five years of professional experience, in a social validation procedure.

STUDY I

METHOD

Participants and Setting

The participants (Andy, Arthur, Benjamin, Charles, Daniel, Eddie, and Tod) were recruited because they met the eligibility criteria, namely (a) a diagnosis of AS, (b) severe to profound developmental disabilities, (c) capacity of manipulating objects, and (d) tongue protrusion, and (e) chronological age comprised between 6 and 10 years (i.e., primary school). Thus, they were diagnosed with AS by their neurologist through DNA results and laboratory tests. The participants were aged of 8.5, 7.3, 9.6, 7.8, 9.7, 10.2, and 8.4 at the beginning of the study, respectively. Their mental ages assessed by their psychologist through Vineland Adaptive Behavioral Scale (VABS; Micheletti et al., 2016) were 1.5, 1.7, 1.8, 2.3, 2.2, 1.6, and 1.8, respectively. Accordingly, they were estimated within the severe to profound range of developmental and intellectual disabilities, although no formal IQ score was available because no test was feasible due to their clinical conditions.

They attended regular classes with a special educational program supervised by their support teacher 24 h per week, focused on promoting both attention and communication skills. They presented lack of speech, stereotypic behavior of hand flapping, tongue protrusion, and drooling. They were reported by their parents and caregivers with a significant amount of hyperactivity during the day. Arthur, Charles, and Tod received speech therapy twice a week. Andy, Benjamin, Daniel, and Eddie were exposed to a stimulation program three days per week. However, they were able of manipulation objects if adequately motivated and rewarded, as described by their parents and caregivers.

The rehabilitative program was implemented individually at participants' homes. It took place in the participants' room. In fact, they were sat in front of a rectangular 120 x 60 cm table. Their families highly appreciated the opportunity for their children to be included in the intervention. They signed, as legal representative of their children, a formal informed consent. The microswitch-cluster

program was additionally approved by a local scientific and ethic committee and was carried out according to Helsinki Declaration and its later amendments.

Selection of Stimuli

Combined with an informal 30 min interview with parents, caregivers, and teachers, a formal preference screening was assessed (Crawford & Schuster, 1993). During the formal screening evaluation, 6 5 min sessions were collected. Auditory, tactile, and visual stimuli to be used as positive reinforcements along the intervention and cluster phases were considered. Overall, 10-15 randomized 10 s of stimuli presentations with a 15 s of rest interval were provided and screened by two research assistants. According to three basic preference criteria, namely (a) alert, (b) orientation, and (c) smiling, stimuli selected by the participants' reactions at least for the 90% of the presentations were retained. Amusing and funny songs, colored lights, familiar voices, and pleasant videos were identified as primary reinforcements for the rehabilitative program.

Technology and Responses

The selected responses consisted of object manipulation (i.e., adaptive responding, which seemed to be the most plausible response for all the participants in accordance with their families and caregivers), and tongue protrusion (i.e., challenging behavior). The adapted microswitches were a wobble microswitch for the adaptive responding and an optic sensor for the challenging behavior. The wobble was a pressure microswitch, ball-like, which should be pulled, pushed, and/or moved left or right side to be activated. The optic sensor was a photocell included in an adapted frame and fixed on the participants' lips corner. A forward tongue protrusion of 1 cm activated the microswitch and was automatically recorded as a challenging behavior by the system. Both combined microswitches constituted the cluster (Stasolla, Perilli, et al., 2014).

The technology additionally included a laptop with a 15.6-inches monitor and a Clicker 5 software package (Crick House, Moulton Park, Northampton, UK). During the sessions, the wobble microswitch was fixed on the table in front of the participants. The computer was behind the wobble, visible but inaccessible to the participants. The rationale of the current responses was to (a) promote object manipulation, (b) enhance constructive engagement, (c) reduce participants' hyperactivity, and (d) decrease tongue protrusion (Stasolla, Perilli, et al., 2017).

Sessions and Data Collection

Sessions lasted 5 min and were video-recorded. Typically, 4 sessions were conducted 3 days per week. The study was performed along 27 months (i.e., a 2-year follow-up included). Overall, 130 sessions were collected for each participant. Because the positive reinforcement lasted 10 s, the participants could provide up to 30 adaptive responses along each 5 min session. The laptop served for the following functions: (a) recording automatically both adaptive responses and challenging behaviors, (b) ignoring a new adaptive response occurring during the stimulation period, and (c) delivering automatically the randomized positive stimulation during intervention and follow-up phases.

Data collection included the following dependent variables: (a) the adaptive responses, (b) the challenging behavior, and (c) the indices of positive participation (i.e., gaze oriented to the wobble and the laptop, smiling, freezing as sign of captured and directed attention to the technology and the rehabilitative program). The indices of positive participation were recorded according to a 15 s partial interval coding system, with 10 s of observation followed by 5 s of dichotomous absence/presence recording of the participation eventually occurred during the previous 10 s interval (Stasolla, Caffò, et al., 2017).

Inter-Observers Agreement

Two blind independent research assistants simultaneously coded the dependent variables on all the collected sessions. The agreement percentages were 99% (range 96-100%), 99% (range 97-100%), and 97% (range 95-100%) for the adaptive responding, the challenging behavior, and the positive participation, respectively. Moreover, Cohen's K scores were .99 for both adaptive responding and challenging behavior, and .97 for positive participation.

Procedural Integrity

The aforementioned blind research assistants followed the study step by step. Thus, both coded the sessions independently according to a provided checklist (i.e., selection of stimuli, data recording, and stimulation delivery). Subsequently, a systematic and simultaneous comparison between both occurred. Finally, they supervised the follow-up and the social validation procedure (i.e., see Study II). A 100% of corrected procedural implementation was empirically evidenced.

Experimental Conditions

The study was carried out according to an ABB^1AB^1 experimental sequence (Barlow, Nock, & Hersen, 2009; Kennedy, 2005). A 2-year follow-up was additionally conducted. A indicated the baselines, B indicated the intervention phase aimed at promoting the adaptive responding irrespective of the challenging behavior, and B^1 indicated the cluster phases.

First Baseline

During the first baseline, 5 sessions were collected within two days for each participant. The technology was available but inactive. No environmental consequences were delivered by the technological system, even if an adaptive responding was exhibited free of the challenging behavior.

Intervention Phase

During the intervention, the technology was available and active. However, this phase was aimed at promoting the adaptive responding irrespective of the challenging behavior. That is, 10 s of positive stimulation was automatically delivered by the system contingently to the exhibition of an adaptive responding regardless of the simultaneous absence or the presence of a challenging behavior. Twenty sessions were collected for each participant along two weeks.

First Cluster

During the cluster, the technology was available and active. An adaptive responding was contingently reinforced by the system with 10 s of positive stimulation only if it occurred free of the challenging behavior. Whenever a challenging behavior was exhibited during the stimulation delivery, the technological system instantly interrupted the delivery itself. Twenty-five sessions were collected along three weeks for each participant.

Second Baseline

To assess the participants' awareness of the cluster program, once the adaptive responding increased and the challenging behavior was significantly reduced, a new baseline occurred. Conditions were identical to the first baseline and five new sessions were collected for each participant along two days.

Second Cluster

Finally, a new cluster phase was implemented. Conditions were identical to the first cluster phase. Twenty-five sessions were collected for each participant along three weeks.

Follow-up

A 2-year follow-up was conducted. Thus, a new double AB[1] experimental sequence was evaluated. Fifty sessions were collected for each participant, equally divided with 5 and 20 sessions for both baselines and cluster phases, respectively. The follow-up lasted one month.

RESULTS

Data were deposited in a public repository (DOI: 10.6084/m9.figshare.12665366), summarized in Tables 1-3 and plotted in Figure 1. The adaptive responding was reported with mean frequencies and ranges. Because the challenging behavior was recorded simultaneously of the adaptive responding, it was reported with a percentage as it represented its portion, beside the mean frequency. Similarly, the intervals of indices with positive participation were indicated with a percentage as well.

INSERT TABLE 1, 2, 3 HERE

INSERT FIGURE 1 HERE

Both descriptive data and graphical inspection revealed an increased adaptive responding and positive participation along the intervention phases for all the participants involved. Conversely, the portion of the challenging behavior was significantly reduced. A series of comparisons using the Tau-U index proposed by Parker, Vannest, Davis, and Sauber (2011) were performed between (a) first baseline and intervention, (b) first baseline and first cluster, (c) second baseline and second cluster,

(d) intervention and first cluster, (e) intervention and second cluster, (f) first follow-up baseline and first follow-up cluster, (g) second follow-up baseline and second follow-up cluster, (h) second cluster and first follow-up cluster, for each participant and for each variable (adaptive responding, challenging behavior, and positive participation). Overall, 168 comparisons were performed, namely eight comparisons on three variables for seven participants. Tau-U effect sizes varied between 0.96 and 0.99 (Median = 0.98), between 0.01 and 0.97 (Median = 0.58), and between 0.96 and 0.99 (Median = 0.98) for the comparisons between baselines and intervention or cluster phases for all the participants and for adaptive responding, challenging behavior and positive participation variables, respectively. They varied between 0.31 and 0.61 (Median = 0.56), between 0.21 and 0.60 (Median = 0.50), and between 0.24 and 0.75 (Median = 0.61) for the comparisons between intervention phase and the first two cluster phases for all the participants and for adaptive responding, challenging behavior and positive participation variables, respectively. They varied between 0.28 and 0.74 (Median = 0.55), between 0.04 and 0.38 (Median = 0.20), and between 0 and 0.61 (Median = 0.41) for the comparison between the second cluster phase and the first cluster phase of the follow-up for all the participants and for adaptive responding, challenging behavior and positive participation variables, respectively.

STUDY II

PARTICIPANTS

Fifty-six external raters with at least 5 years of professional experience were involved in a social validation procedure. They were equally divided into 4 groups. Specifically, 14 among caregivers, psychologists, physiotherapists, and teachers. Their mean ages were 38.57, 41.78, 46.21, and 40.71 and related standard deviations were 2.79, 4.17, 2.39, and 3.24, for caregivers, psychologists, physiotherapists, and teachers, respectively. They represented a convenience sample of professionals

interested in the field of developmental disabilities. They were eligible because, added to their professional experience above indicated, they supervised at least one AT-based intervention in children with developmental disorders. The pool from which the raters were recruited included (a) parents of children with developmental disabilities, (b) neurologists, (c) psychologists, (d) physiotherapists, (e) speech therapists, (f) behavior analysts, and (g) teachers. The participants were completely blind to the research goals. Both parents and caregivers of the participants enrolled in Study I were preliminarily excluded to avoid biased evaluations.

PROCEDURE

They were told that they were requested to watch a 8-min video including a child with AS and developmental disabilities exposed to an AT-based intervention, in 4 different experimental conditions. They were divided in small groups of 4 raters. Each participant was randomly assigned to 2 groups (i.e., eight raters). Raters watched one of the participants included in Study I in a randomized sequence of 4 experimental conditions, (a) baseline, (b) intervention, (c) cluster, and (d) follow-up. Each video represented a 2-min standard session of the aforementioned conditions. Each experimental condition was detailed and explained by a research assistant as reported in the experimental conditions sub-heading of Study I. Raters were invited to fill an 8-item questionnaire (see Table 4), which was validated by Stasolla et al. (2019), and each item was rated on a 5 point Likert-like scale, which varied from totally disagree (=1) to totally agree (=5).

INSERT TABLE 4 HERE

STATISTICAL ANALYSIS

Statistical analysis was performed with R software. A general linear model was employed to test the null hypothesis that there were no differences across conditions (i.e. baseline, intervention, cluster, and follow-up phases), and across the groups of raters (i.e. professions: caregiver, physiotherapist, psychologist, and teacher) for the seven children involved in Study I, along the eight items of the questionnaire. The general linear model was chosen in order to control for each independent and dependent variable included in the analysis. Follow-up analyses were conducted, as appropriate, to test for significant main and first order interaction effects on univariate measures. A p-value $< .05$ was determined to be significant for interpreting multivariate results, and a p-value $< .0071$ was determined to be significant for interpreting univariate results, as effect of a Bonferroni correction, i.e. the initial alpha level (.05) was divided for the number of dependent variables (6). The present analysis was conducted in order to formally endorse the use of the AT by expert external raters.

RESULTS

Data were deposited in a public repository (DOI: 10.6084/m9.figshare.12665393). Results of the general linear model showed that the main multivariate effects of condition (Wilks' Lambda = 0.096, $F(3,208) = 30.26$, $p < 0.001$) and groups of raters (Wilks' Lambda = 0.779, $F(3,280) = 2.19$, $p < 0.05$) were significant. Raters scored more favorably cluster (M = 4.46, SD = 0.48) and follow-up (M = 4.46, SD = 0.48) sessions with respect to intervention (M = 3.23, SD = 0.38) and baseline (M = 1.51, SD = 0.46) sessions. Psychologists (M = 3.60, SD = 1.47) scored more favorably the video-clips with respect to caregivers (M = 3.39, SD = 1.30), physiotherapists (M = 3.36, SD = 1.19), and teachers (M = 3.31, SD = 1.22). First-order interaction effect was not significant. Regarding univariate effects, the score differences between baseline, intervention and cluster sessions were statistically significant ($p < 0.001$) for all the eight items of the questionnaire, clearly favoring cluster sessions over baseline and intervention sessions. No differences were found between cluster and follow-up sessions. The score differences among the four groups of raters were statistically significant for five out of eight

items (at least with $p < 0.05$), namely rehabilitation outcomes, positive participation, home and school contexts, agreement with the condition. Regarding rehabilitation outcomes and positive participation psychologists scored more favorably than the other three groups; regarding home and school contexts, and agreement with the condition psychologists scored more positively than physiotherapists and teachers. Table 5 reports mean scores and standard deviations on the eight questionnaire items for the four conditions and for the four groups of raters.

INSERT TABLE 5 HERE

GENERAL DISCUSSION

Data of both studies further extended and confirmed the effectiveness and the suitability of the MCT to promote an adaptive responding and reduce simultaneously a challenging behavior among pediatric populations affected by severe to profound developmental disabilities. The current investigation represented a favorable research extension within this framework with regard to (a) new participants with AS, (b) a long-term follow-up (i.e., 24 months), and (c) new groups of raters and experimental conditions observed along the social validation procedure. Results demonstrated that all the participants profitably learned the functional use of the technology to enhance object manipulation, decrease tongue protrusion, and positively participated to the sessions, being constructively engaged with an active role. All the participants consolidated the learning process over two years. Social raters favorably evaluated the use of the technology and corroborated the intervention's social and clinical validity accordingly. Data were supported by previous findings (Lancioni, O'Reilly, et al., 2011, 2013; Lancioni, Singh, O'Reilly, Sigafoos, & Oliva, 2014; Lancioni, Singh, O’Reilly, Sigafoos, Oliva, et al., 2007; Lancioni, Smaldone, et al., 2007) and suggested some considerations.

First, both studies may be viewed as a research extension of previous findings aimed at fostering an adaptive responding and simultaneously decreasing a challenging behavior through a

MCT among pediatric populations with severe to profound developmental disabilities. The current investigation was the first attempt available in the literature to expose children with AS and multiple disabilities to a MCT helpful to promote independence and self-determination of the participants involved. MCT ensured the participants with the autonomous access to pleasant environmental events. Thus, the participants first learned to produce a new adaptive response for manipulating object and subsequently reduced their challenging behavior during cluster phases. Similarly, they favorably participated and improved their active role towards their environmental stimuli preventing their hyperactivity, with beneficial consequences on their social desirability, image, and status since they were constructively engaged (Chadwick & Platt, 2018; Stasolla & De Pace, 2014; Wheeler, Sacco, & Cabo, 2017).

Second, MCT may be conceived as a valid solution with regard to conventional programs (e.g., non-contingent reinforcement and/or response cost) for reducing caregivers' burden. It may represent a crucial educational and rehabilitative option for improving the participants' inclusion in daily settings. In fact, MCT can be adopted as a relative realistic and/or practical program for serious and difficult clinical conditions. That is, the simultaneous monitoring of two dependent variables (i.e., both adaptive responding and challenging behavior), the delivery of contingent positive events, and the interruption of it whenever the challenging behavior occurred, may be precarious and/or heavy for families and caregivers to be practiced. Consequently, he MCT can meet all the above listed requirements. Its application may be considered as affordable by families and caregivers (informally interviewed) in daily context since its cost is approximately 850 US dollars (Boot et al., 2018; Chadha, Moussy, & Friede, 2014).

Third, the success of the MCT used in the current investigation was largely based on the rewarding value of the contingent pleasant events delivered, the simplicity (i.e., low cost of the adaptive responding because already available in the individual repertoire), the manageability of the challenging behavior, and the practicality of the adopted devices. With regard to the first point, strong

(i.e., relevantly motivating) positive stimulation may adequately supply the response cost. With regard to the second point, an existing and easily reproducible adaptive response to be performed may be viewed as mandatory for the learning process. With regard to the third point, the monitoring of the challenging behavior widely relied on the rapidity with it may be redirected and/or substituted by a new adaptive responding. With regard to the fourth point, the effectiveness of the technological options selected can be evaluated with the easy association between the adopted microswitches and the targeted responses/behaviors (Lancioni, Sigafoos, O'Reilly, & Singh, 2012; Lancioni, Singh, O'Reilly, Sigafoos, Chiapparino, et al., 2007). The empirical data collected along the current investigation confirmed a positive outcome on each of the aforementioned features.

Fourth, the favorable learning acquired by the participants through the sessions of the study was positively consolidated over the time. Thus, the long-term follow-up (i.e., 24 months) corroborated that all the participants maintained their learned skills for manipulating objects. Moreover, they significantly decreased their tongue protrusion. One may argue that their cognitive and behavioral capacities were fostered, even with a long-term intervention interruption (i.e., two years), confirming the validity of the MCT for rehabilitative purposes (Catania, 2012; Kazdin, 2001; Stasolla, Damiani, et al., 2014).

Fifth, the social raters positively evaluated the implementation of MCT for promoting independent manipulation of objects, decreasing tongue protrusion, and improving positive participation. Irrespective of their group (i.e., caregivers, physiotherapists, psychologists or teachers), they all assessed the cluster option suitable for the dual goal of learning an adaptive responding and simultaneously reducing a challenging behavior. Essentially, both clinical and social validity of the rehabilitative program was externally confirmed (Lancioni, O'Reilly, et al., 2006; Stasolla, Damiani, et al., 2015). Some differences between the four groups emerged for items regarding rehabilitation outcomes, positive participation, and suitability for different contexts, whereas a trend emerged for psychologists to rate more favorably than other groups such items. This could reflect different degrees

of specific competence as well as of involvement in daily practices for each group, but all of them substantially agreed in considering the MCT program effective, suitable and affordable.

LIMITATIONS

Despite the above argued enthusiastic results, this investigation presented some limitations. For instance, it was based on a single-subject experimental design, even if the small sample included was representative. Caution was mandatory accordingly, and its generalization to new participants with AS and/other rare genetic syndromes (e.g., Fragile X, Cornelia de Lange, and Rett) was recommended. Additionally, different adaptive and functional responses (e.g., sorting objects) could be considered. Moreover, a functional assessment of the challenging behavior would be helpful (Falcomata & Lang, 2013). The current investigation was based on simple responses. Different and more complex behavioral responses could be learned for the future. A systematic comparison with other cognitive-behavioral interventions (e.g., extinction and response cost) would be helpful (i.e., one managed via microswitch and one monitored via caregiver).

FUTURE RESEARCH PERSPECTIVES

In light of the above, future research within this framework should tackle the following points (a) new extension of the MCT to further participants with AS and/other developmental disorders (e.g., autism and/or cerebral palsy), (b) generalization of its use in daily environments (e.g., school and/or rehabilitative settings), (c) a systematic comparison with different AT-based programs (e.g., choice between different categories of stimuli) or other cognitive-behavioral interventions (e.g., differential reinforcement of an alternative behavior), (d) a further extension of the social validation procedure to new groups of raters (e.g., parents of children with developmental disabilities, and/or neurologists),

and (e) the inclusion of preference checks for the participants involved between two different rehabilitative interventions adopted.

Compliance with Ethical Standards

Declaration of conflicts of interest

The authors declared no conflicts of interest with respect to the research, authorship, and/or publication of the article. The authors alone are responsible for the content and the writing of the article.

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Informed Consent

Informed consent was obtained for all the recruited participants by their legal representatives (i.e., their parents).

Ethical Approval

All performed procedures of the study have been carried out in accordance with Helsinki Declaration (1964) and its later amendments or comparable ethical standards.

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Table 1. Mean frequencies and ranges of adaptive responses.

Participants	Intervention phases					Follow up			
	A	B	B'	A	B'	A	B'	A	B'
Andy	2.6 (2-4)	17.5 (8-22)	22.4 (20-26)	5.4 (4-7)	22.64 (19-24)	3 (2-5)	17.95 (14-21)	4 (2-6)	19.65 (17-24)
Arthur	2.8 (2-4)	15.5 (11-20)	23.92 (21-26)	3.4 (2-5)	23.64 (20-26)	2 (1-3)	20.1 (16-24)	3.4 (2-5)	17.7 (17-24)
Benjamin	2.8 (2-5)	16 (12-18)	21.88 (20-24)	2.8 (2-4)	21.4 (14-24)	2.4 (2-3)	19.8 (12-24)	3.2 (2-5)	20.65 (14-24)
Charles	2.2 (1-3)	16.95 (11-21)	24.72 (22-28)	4.2 (1-6)	24 (18-28)	3.2 (2-5)	21.35 (16-26)	3.8 (2-8)	21.35 (18-24)
Daniel	3.2 (2-4)	19.15 (12-24)	25.44 (23-28)	3.2 (2-6)	23.48 (13-29)	2.8 (2-5)	19 (12-25)	3.6 (1-5)	23.25 (19-26)
Eddie	5 (4-6)	15.1 (10-18)	22.36 (20-25)	3.4 (2-5)	23.04 (18-26)	2 (1-3)	15.1 (11-18)	2 (1-4)	16.45 (14-18)
Tod	5.6 (4-8)	15.65 (12-18)	23.6 (21-26)	5.4 (3-7)	22.28 (15-26)	2 (1-4)	18.9 (17-22)	3 (2-5)	17.65 (13-20)

Table 2. Mean percentages, ranges, and mean frequencies of challenging behaviors.

Participants	Intervention phases					Follow up			
	A	B	B'	A	B'	A	B'	A	B'
Andy	100 (100-100) - 2.6	39.61 (25-75) - 6.35	10.80 (4-17) - 2.52	94.28(71-100) - 5	7.67 (4-9) - 1.72	96 (80-100) - 2.8	10.54 (5-20) - 1.85	96.66(83-100) - 3.8	8.91 (4-20) - 1,7
Arthur	100 (100-100) - 2.8	43.94(26-92) - 6.5	11.42(4-26) - 2,72	100 (100-100) - 3.4	7.74 (4-5) - 1.8	100 (100-100) - 2	9.52 (4-19) - 1.85	100 (100-100) - 3.4	9.84 (5-13) - 1.7
Benjamin	100 (100-100) - 2.8	31.64 (4-11) - 4.85	8.250 (4-13) - 1.8	100 (100-100) - 2.8	8.52 (4-13) - 1.68	100 (100-100) - 2.4	12.51 (5-33) - 2.35	100 (100-100) - 3.2	8.82 (4-13) - 1.75
Charles	100 (100-100) - 2.2	26.93 (9-73) - 4.2	11.52(4-23) - 2.84	96.66 (83-100) - 4	8.26 (4-17) - 1.96	100 (100-100) - 3.2	10.93 (4-24) - 2.25	100 (100-100) - 3.8	9.05 (4-11) - 1.9
Daniel	100 (100-100) - 3.2	18.82 (5-75) - 3.2	6.96 (4-17) - 2.52	100 (100-100) - 3.2	7.22 (3-23) - 1.56	100 (100-100) - 2.8	11,50 (4-58) - 1.85	100 (100-100) - 3.6	6.38 (4-16) - 1.45
Eddie	96.66(83-100) - 4.8	17.85 (11-36) - 2.55	5.55 (0-9) - 1.24	100 (100-100) - 3.4	3.70 (0-9) - 0.84	100 (100-100) - 2	9.98 (6-27) - 1.45	100 (100-100) - 2	7.69 (5-13) - 1.25
Tod	96.66 (83-42) - 5.4	18.17(11-42) - 2.75	3.25 (0-9) - 0.76	97.14 (86-100) - 5.2	5.28 (0-20) - 1.12	100 (100-100) - 2	8.24 (5-17) - 1.55	100 (100-100) - 3	6.72 (0-15) - 1.15

Table 3. Mean percentages and ranges of intervals with indices of positive participation.

Participants	Intervention phases					Follow up			
	A	B	B'	A	B'	A	B'	A	B'
Andy	18 (10-25)	64.5 (50-75)	78.37 (70-90)	29 (10-20)	80.8 (65-90)	18 (10-25)	78.25 (65-90)	20 (15-25)	79.75 (70-90)
Arthur	25 (20-30)	65.5 (55-80)	88 (70-100)	28 (20-40)	86.2 (70-100)	21 (10-30)	70.5 (60-80)	14 (10-20)	76 (60-90)
Benjamin	22 (20-25)	64.5 (55-75)	81.8 (65-100)	14 (10-20)	79.8 (60-100)	16 (10-30)	69.5 (60-80)	8 (5-10)	72.75 (60-85)
Charles	26 (20-30)	69 (60-80)	83.4 (65-100)	20 (10-35)	74.2 (60-90)	24 (20-30)	76.5 (65-90)	21 (10-25)	72.25 (60-90)
Daniel	24 (20-30)	68 (65-75)	83.6 (70-100)	26 (20-35)	80.2 (70-90)	18 (10-30)	74.25 (65-85)	21 (15-25)	77 (65-90)
Eddie	31 (25-35)	67.5 (55-75)	84.6 (75-90)	22(10-35)	80.2 (70-90)	19 (15-25)	80.75 (70-90)	22 (10-40)	78.5 (60-90)
Tod	33 (25-40)	68.75 (60-80)	88.2 (70-100)	30 (15-40)	84.4 (70-95)	15 (10-20)	68.75 (60-80)	12 (5-25)	76.75 (65-90)

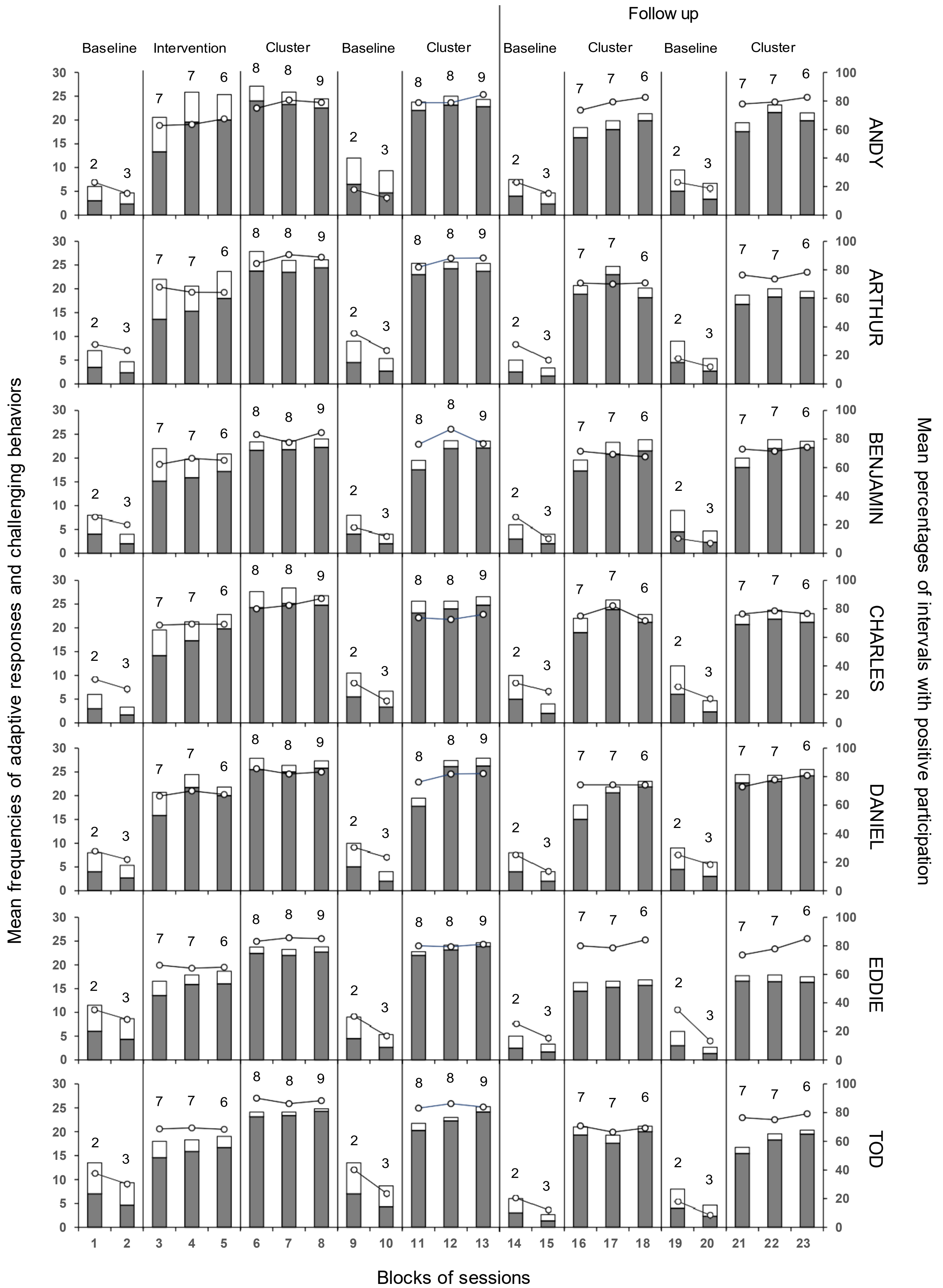
Table 4. Social validation questionnaire

Do you think the child enjoys (is comfortable with) this condition?
Do you think this condition promotes self-determination?
Do you think this condition has beneficial/rehabilitative outcomes?
Do you think this condition enhances positive participation?
Do you think this condition is suitable for home contexts?
Do you think this condition is suitable for school contexts?
Do you think this condition is suitable for medical/rehabilitative contexts?
Do you support (agree with) this condition?

Table 5. Mean scores and standard deviations on the eight questionnaire items for the four conditions and for the four groups of raters.

Condition	Comfort	Self Determination	Rehabilitation	Participation	Home Context	School Context	Medical Context	Agreement
Baseline	1.59 ± 0.56	1.52 ± 0.57	1.50 ± 0.57	1.55 ± 0.57	1.50 ± 0.57	1.52 ± 0.57	1.45 ± 0.57	1.41 ± 0.56
Intervention	3.21 ± 0.46	3.25 ± 0.44	3.27 ± 0.45	3.23 ± 0.43	3.23 ± 0.43	3.21 ± 0.41	3.23 ± 0.43	3.20 ± 0.44
Cluster	4.45 ± 0.50	4.45 ± 0.50	4.52 ± 0.50	4.48 ± 0.50	4.45 ± 0.50	4.45 ± 0.50	4.45 ± 0.50	4.45 ± 0.50
Follow_up	4.45 ± 0.50	4.45 ± 0.50	4.52 ± 0.50	4.48 ± 0.50	4.45 ± 0.50	4.45 ± 0.50	4.45 ± 0.50	4.45 ± 0.50

Profession	Comfort	Self Determination	Rehabilitation	Participation	Home Context	School Context	Medical Context	Agreement
Caregiver	1.30 ± 3.41	1.30 ± 3.41	1.33 ± 3.36	1.33 ± 3.38	1.33 ± 3.38	1.28 ± 3.41	1.30 ± 3.41	1.33 ± 3.38
Physiotherapist	1.22 ± 3.45	1.25 ± 3.43	1.25 ± 3.43	1.12 ± 3.36	1.20 ± 3.30	1.20 ± 3.30	1.18 ± 3.32	1.23 ± 3.29
Psychologist	1.39 ± 3.57	1.46 ± 3.52	1.50 ± 3.68	1.48 ± 3.68	1.49 ± 3.61	1.51 ± 3.59	1.56 ± 3.55	1.51 ± 3.57
Teachers	1.21 ± 3.27	1.20 ± 3.30	1.27 ± 3.34	1.27 ± 3.34	1.21 ± 3.34	1.21 ± 3.32	1.26 ± 3.29	1.30 ± 3.27



Follow up
Baseline
Intervention
Cluster
Baseline
Cluster
Baseline
Cluster
Baseline
Cluster
ANDY
ARTHUR
BENJAMIN
CHARLES
DANIEL
EDDIE
TOD
Mean frequencies of adaptive responses and challenging behaviors
Mean percentages of intervals with positive participation
Blocks of sessions

Figure 1. The graph summarizes the data for the seven participants. The dark grey and the light bars indicate the mean frequencies of adaptive responses and challenging behaviors, respectively, over blocks of sessions for the baseline, intervention and cluster phases and the follow-up phases. The white dots refer to the mean percentage of intervals with positive participation over blocks of sessions for the baseline, intervention and cluster phases and the follow-up phases. The number of sessions included in each block is indicated by the numeral above it.