Improving Theory of Mind skills in Down Syndrome? A pilot Study.

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Abstract

Effective communication requires an understanding of the interlocutor's perspective. Being able to infer someone else's knowledge about a situation is a critical skill in any communication and social interaction. These abilities are part of Theory of Mind (ToM) skills and are known to be impaired in Down Syndrome (DS). It therefore makes sense to investigate ToM development in this population. In our pilot study, we explore the possibility of improving ToM abilities in participants with DS and typically developing children (TD) matched for nonverbal mental age. Participants were assessed with the French adaptation of the "ToM Inventory" before and after a 10-week training session. Results show that trained groups perform significantly better on ToM tasks than untrained groups, whose performances remain stable between pre- and post-test. These results are encouraging as they suggest that, with a specific training, children with DS can improve their ToM skills.

Keywords: Down Syndrome, Theory of Mind; Theory of Mind Training, Social cognition

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INTRODUCTION

Theory of Mind (ToM) includes the ability to attribute mental states (thoughts, emotions, beliefs, desires, etc.) to others, as well as the ability to admit that others' thoughts or feelings may differ from ours (Losh, Martin, Klusek, Hogan-Brown, & Sideris, 2012). This knowledge is essential to explain and predict someone else's behavior as well as to communicate efficiently and more specifically to acquire the pragmatics of language (Losh et al., 2012). Being able to understand someone else's perspective and to infer his/her knowledge about a situation is a critical skill in any communication and social interaction. The necessary abilities for the emergence of ToM skills develop between the age of 2 and 5-year-old. Generally basic ToM skills come to maturity around the age of 4 or 5-year-old in typically developing children. ToM skills do not develop in the same way for all children. It is more common for children with genetic or neurodevelopmental disorders to have difficulties with ToM tasks. The first studies on pathological populations focused on autism, a neurodevelopmental disorder characterized by a notable deficit in social interactions (Baron-Cohen, 1989; Baron-Cohen, Leslie, & Frith, 1985). While these pioneering studies considered the ToM deficit as specific to autism, subsequent studies found similar deficits in other neurodevelopmental disorders such as intellectual disability (ID) (see Cobos & Castro, 2010 for a review). In ToM studies, autistic subjects are generally the target group and children with ID are considered as a control group so that few researches have been conducted on individuals with ID per se (Giouri, Alevriadou, & Taskiridou, 2010). However, in addition to a significant limitation in intellectual functioning, individuals with ID are also characterized by limitations in adaptive behavior including everyday social skills (American Association Intellectual and Development Disabilities [AAIDD], 2010). ToM skills are at the intersection of these two areas as they require an adequate level in both intellectual functioning and adaptive behavior. It is therefore interesting to better understand the impact of intellectual disability, which is the most common cause of intellectual disability (10% of all cases in the absence of any prenatal screening). Its prevalence is estimated at 1/750-2000 live births depending on countries' religious and sociocultural practices (Diamandopoulos & Green, 2018). Even if DS is the most studied genetic syndrome of intellectual disability (Touraine, de Freminville, & Sanlaville, 2011), some aspects of cognitive and linguistic development remain poorly documented (e.g. lexical and semantic development, categorization strategies, referential communication and more broadly social communication and social cognition) (see Comblain & Thibaut, in press, for a review). Individuals with DS generally fail to perceive someone else's mental states and tend to assign their own thoughts to others. The literature on false beliefs and emotion recognition in DS points to a deficit in these areas. Moreover, ToM abilities appear to be quite poor in individuals with DS (Thirion-Marissiaux & Nader-Grosbois, 2008; Nader Grobois, 2011) whose performance in traditional ToM tasks is lower than that of peers with non-specific ID and typically developing children (TD) (Giaouri et al., 2010). Very few studies focus on ToM in individuals with DS, and usually highlight a developmental trajectory qualitatively similar to that of TD children. The difference between children with DS and TD peers tends to rather be in the developmental timing than in ToM stages per se. In other words, the emergence of ToM would be delayed in children with DS compared to TD children (Thirion-Marissiaux & Nader-Grosbois, 2007, 2008). According to Giaouri et al. (2010) ToM deficits in DS can be partially explained by underlying deficits in skills such as language, executive functions, short-term memory, inhibition, metacognition and attention.

Since the ability to understand someone else's perspective and knowledge is essential to any communication and social interaction, the question of the effectiveness of a specific training in pathological populations is crucial. Few studies address this question and most of these concern TD children (Melot & Angeard, 2003; Westra & Carruthers, 2017). Regarding interventions in DS we only noted a case study of a 24-year-old adult (Montoya-Rodriguez, McHugh, Cobos, 2017) focusing on words expressing deictic relations ("I", "you", "he", "there", "now" and "then"). Studies on ToM training in TD children are more frequent. We will briefly discus three of them. In 2003, Melot and Angeard conducted a study on 111 TD children aged 3;6 to 4;4 years old focusing on false-belief prerequisites. They showed that training these prerequisites through appearance-reality and false-belief activities improves performance in false-belief tasks. Moreover, it seems that, on the one hand, performance is specific to training and, on the other hand, remains stable over time. More recently, Westra and Carruthers (2017) conducted a study on 75 TD children aged 3 to 5 years old. Various tasks involving the training of desire, beliefs, hidden emotions, knowledge and false beliefs were presented to children. The impact of training on belief and false-belief tasks was different: it was greater regarding beliefs than on false beliefs, the latter requiring pragmatic skills not yet developed in such young children. The authors suggested that a specific training lasting 6 to 12 months is necessary to observe an improvement in false-belief tasks. Moreover, mental states understanding, questions to be asked to elicit a specific situation as well as distinction between relevant and irrelevant cues can also be improved by training. These improvements probably reflect the child's new ability to interpret mental states (Westra & Carruthers, 2017). However, no generalization of the effects of training on other skills was observed. In 2016, Gombert, Bernat and Roussey studied the progress of 65 TD children aged 5, trained with a multimodal method using visual, audio-visual, kinesthetic, olfactory material as well as books in a shared reading activity. Gombert et al.'s (2016) results highlight a significant improvement in ToM abilities in the trained group compared to the control group. In addition, the authors stress that using books helps to stimulate imagination and thus to train the ability to infer the knowledge, feelings and emotions of the characters. This procedure promotes an active participation of the child that could be worth developing among children with Down syndrome.

GOALS AND HYPOTHESES

The main objectives of this pilot study are: (1) to compare the Theory of Mind (ToM) level of individuals with DS and control TD children matched for nonverbal mental age and (2) to evaluate the effectiveness of a specific training program on ToM tasks in both groups. Our hypotheses are: (1) according to the delay hypothesis (Thirion-Marissiaux & Nader-Grosbois, 2008), participants with DS will perform less well than TD children in Theory of Mind tasks (ToM Inventory), and (2) according to Gombert et al.'s results, both experimental groups (5 trained SD participants and 5 trained TD children) will have higher ToM scores in the post-test than in the pre-test, while both control groups (5 untrained SD participants and 5 untrained TD children) will have equivalent ToM scores in both post- and pre-tests.

METHOD

Participants

Twenty participants took part in the study: 10 participants with DS (4 females and 6 males, 8.5 to 18.3-year-old, mean 11.5) and 10 TD children (3 females and 7 males, 3.11 to 4.8-year-old, mean 4.3) matched for nonverbal mental age measured with the Raven's "Coloured Progressive Matrix" (Raven, 1998). The French version of the Peabody Picture Vocabulary Test (PPVT) (EVIP, Dunn, Dunn & Theriaults-Whale, 1993) was also administered to all the participants. The statistical analyses were performed with Statistical Package for the Social Sciences (SPSS, 2017). Results are significant at $p < 0.05$. Given our very small sample size, nonparametric statistics are used. DS and TD groups characteristics are summarized in Table1 (ages are expressed in months).

Table 1.
Characteristics of the DS and TD Groups

	Down Syndrome (DS)	Typically Developing Children (TD)
N	10	10
Mean NV-MA	55.20 (SD: 5.33) Range: 48 – 63	57.30 (SD: 9.10) Range: 48 – 75
Mean LexA	53.00 (SD: 13.77) Range: 30 – 75	52.65 (SD: 9.26) Range: 35 – 65

NV-MA = nonverbal mental age, LexA = lexical (age in months)

A Mann-Whitney analysis for independent samples shows that groups were equivalent in nonverbal mental age ($U = 45.50$, $p = 0.74$) and lexical age ($U = 46.50$, $p = 0.80$). Half of the DS and TD participants were randomly assigned to an experimental group and the other half to a control group (Table 2).

Table 2.
Characteristics of the Experimental and the Control groups

	Down Syndrome		Typically Developing Children	
	Experimental Group (DS-Experimental)	Control Group (DS-Control)	Experimental Group (TD-Experimental)	Control Group (TD-Control)
N	5	5	5	5
Mean NV-MA	55.20 (SD: 6.22) Range: 48 – 63	55.20 (SD: 5.02) Range: 48 – 60	60 (SD: 11.02) Range: 48 – 75	54.6 (SD: 6.84) Range: 48 – 63
Mean LexA	40.40 (SD: 14.12) Range: 21-61	50.20 (SD: 13.20) Range: 29-69	47.40 (SD: 11.64) Range: 26-58	41.40 (SD: 6.30) Range: 30-46

NV-MA = nonverbal mental age, LexA = lexical (age in months)

To control that nonverbal mental age and lexical age remained equivalent once divided into subgroups, we used a Kruskal-Wallis analysis [respectively for NV-MA and LexA: $H(3) = 0.84$, $p = 0.84$ and $H(3) = 3.65$, $p = 0.30$].

The participants assigned to the experimental group received a 10-week ToM training while the control participants did not receive any specific training. All the participants were presented twice with ToM tasks: before and after the training sessions (Table 2).

Ethics approval

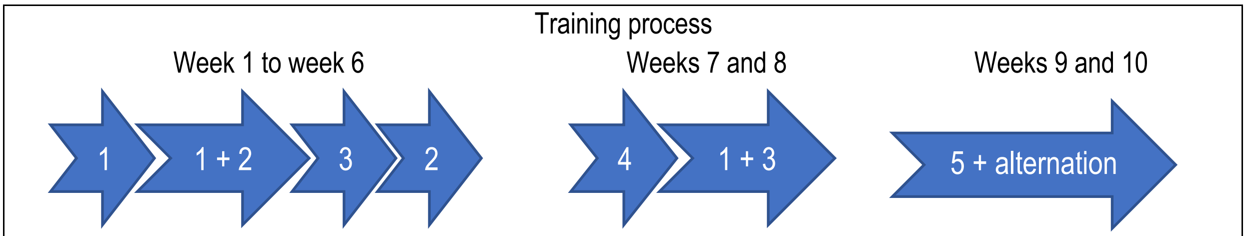
This study was conducted with the approval of the Ethics Committee of the Faculty of Psychology, Logopedics and Educational Sciences of the University of Liège (Liège, Belgium). Each participant was tested by a speech and language pathology student under the supervision of a qualified professional. All the children who took part in the study following had the written consent of their parents. An adapted consent form (with pictograms) was also proposed to all the participants. All the data was anonymized and kept confidential.

Experimental tasks

The experimental protocol was presented twice to all the participants: at the beginning and at the end of the study (immediately after the training process in the experimental group - see experimental design below). All the participants were assessed with the "ToM Inventory" designed by Hutchins, Prelock and Bonazinga-Bouyea (2014) and adapted in French by Nader-Grosbois and Houssa (2016). We chose the "Theory of Mind Inventory" because of the inadequacy of traditional ToM tasks for children with DS. Indeed, one limitation of the traditional ToM tasks is that children's cognitive and language levels can influence performance; therefore, it would be difficult to use them with intellectually impaired individuals such as children with DS (Charman, Campbell & Edwards, 1998). With the "Theory of Mind inventory", it is possible to overcome the difficulties identified in the traditional tasks. It evaluates a wide range of theory of mind competencies and does not suffer from ceiling effects when administered to individuals with linguistic, cognitive, and motivational deficiencies (Hutchins et al., 2014). The protocol was divided into 9 subtests for a total of 15 points. The tasks assess the understanding of emotions, the perception of someone else's desire or emotion as well as beliefs and false beliefs (see Appendix 1 for details).

Training process

Training sessions were conducted on an individual basis once a week over a 10-week period (Figure 1). We focused on identification of emotions and false-belief prerequisites. Five activities were proposed: (1) emotion assignment, (2) shared reading, (3) symbolic game (acting out), (4) referential communication tasks (as designed by Glucksberg, Kauss, & Weisberg, 1966) and (5) emotion assignment (see Appendix 2 for details).



1 = emotion recognition – 2 = shared reading – 3 = symbolic game – 4 = referential communication – 5 = emotion assignment – alternation = alternating previous exercises

Figure 1. Training process

Results

Due to the small sample size, we used nonparametric statistical tests. The signification level is $p < 0.5$. Group data are summarized in Table 3 and Figure 2.

Table 3.
Performance of Experimental and Control Groups on ToM Pre- and Post-test.

		Pretest				Post-test			
Groups	N	Mean	Standard Error	Standard Deviation	Median	Mean	Standard Error	Standard Deviation	Median
DS-Experimental	5	4.20	1.02	2.28	4.00	9.00	0.55	1.22	9.00
DS-Control	5	3.60	0.51	1.14	4.00	3.20	0.58	1.30	3.00
TD-Experimental	5	6.60	0.75	1.67	7.00	13.80	0.58	1.30	14.00
TD-Control	5	7.00	0.55	1.22	7.00	6.40	0.60	1.34	7.00

A Kruskal-Wallis analysis was performed to compare the performance of the experimental and control groups (N = 20) at pre-test and post-test. The results point to a group effect both at pre-test [median: 5.00; $\chi^2(3) = 10.3$, $p = 0.02$] and post-test [median: 13.6; $\chi^2(3) = 13.6$, $p = 0.004$]. Additional Mann-Whitney U tests for independent samples show that the DS control group has significantly lower ToM scores than TD groups both at pretest and post-test (respectively for the pre-test and the post-test when compared with the experimental TD group: U = 2.00, p = 0.03 and U = 0.00, p = 0.009; when compared with the control TD group: U = 0.50, p = 0.01 and U = 1.00, p = 0.14). There is no significant difference between the DS experimental group and TD groups at the pre-test (respectively when compared with the TD experimental group and the TD control group: U = 5.0, p = 0.15 and U = 4.00, p = 0.09). This lack of difference between the DS experimental group and the two TD groups in pre-test is surprising. We expected, as observed in the DS control group, lower performance in the DS experimental group. This is probably due to the uncontrolled intrinsic characteristic of the experimental group. However, if we perform a Mann-Whitney analysis to compare the performance of all DS participants with those of typically developing children, we find a significant difference between the two groups (U = 11.50, p = 0.02) showing that DS participants' ToM scores are lower than those of TD children.

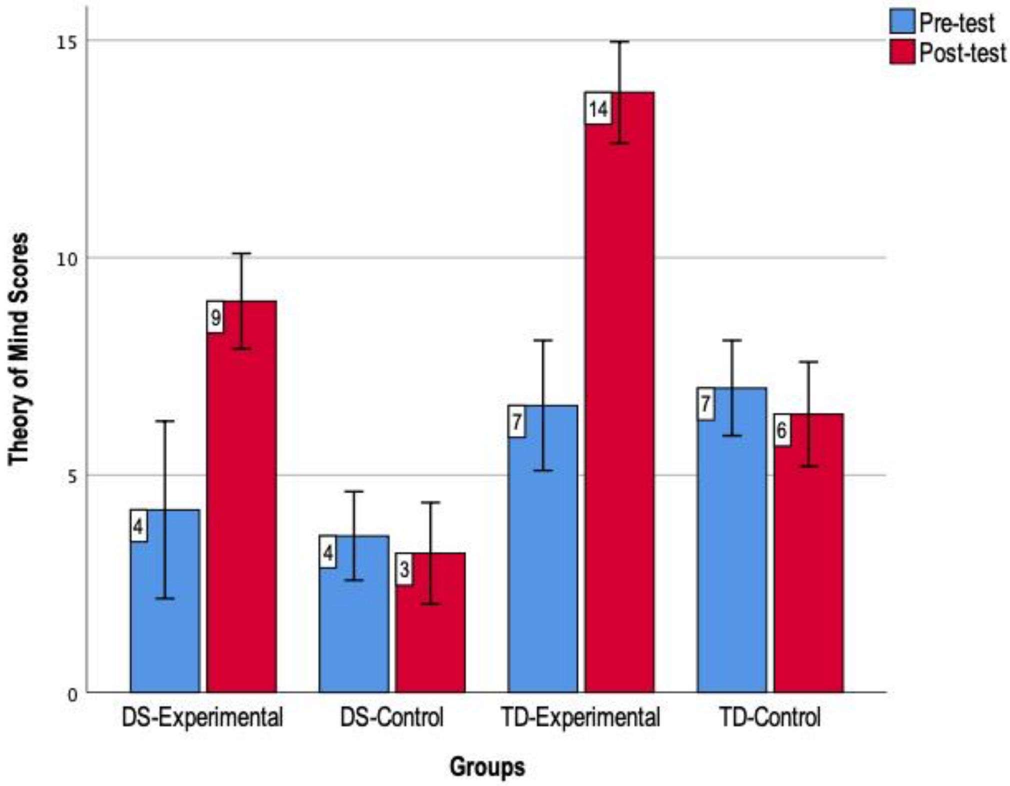


Figure 2. Mean Theory of Mind scores at pre- and post-test

Results summarized in Figure 2 suggest that both experimental groups benefit more from the intervention than the control groups whose performance seems to remain stable. To test this hypothesis and the effect of training in each group, we conducted independent Wilcoxon signed-rank tests for linked samples. Pre- and post-test results were compared for each group. Analyses confirm that both experimental groups have significantly higher ToM scores at post-test than at pretest (DS experimental group: $Z = -2.03$, $p = 0.04$ and TD experimental group: $Z = -2.06$, $p = 0.39$) while the control groups have the same pre-test and pot-test ToM scores (DS control group: $Z = -0.82$, $p = 0.41$ and TD control group: $Z = -0.82$, $p = 0.41$).

Discussion

Before discussing our results, it is important to remember that this study is a pilot study conducted on a very small sample. Therefore, while the results are encouraging, they must be interpreted with caution. The generalization of our conclusions to all the DS population can only be considered after having replicated our results on a larger sample. Moreover, we cannot exclude that the progress made in the post-test is the consequence of the Hawthorne effect. In other words, progress could, at least partially, be attributed to the fact that participants are conscious of participating in an experiment in which they are observed. This awareness of the testing situation can result in greater motivation for the task itself. So, to further strengthen the results, it would also be interesting to add to the initial design a group of DS participants and a group of TD children receiving a substitute intervention.

Our first objective was to compare the overall "Theory of Mind Inventory" scores of participants with Down syndrome with those of typically developing peers matched for NVMA. According to the delay hypothesis (Thirion-Marissiaux & Nader-Grosbois, 2008), we expected participants with DS to perform less well than TD children matched for NVMA. If we consider the DS groups together, our results confirm this hypothesis. DS participants' ToM skills are inferior at pre-test to those of typically developing children matched for nonverbal mental age.

As predicted by our second hypothesis, the two experimental groups (trained DS and TD participants) improved their ToM performances at the end of the training. Our results are therefore in line with those obtained by Gombert et al. (2016). These authors showed a significant improvement in ToM abilities in a group of TD children who had undergone training including shared reading and activities on mental state recognition and emotion assignment.

A qualitative analysis of DS participants' performance is also interesting. In accordance with the developmental trajectory described by Flavell (1999), our subjects seem to understand emotions better than beliefs and to understand beliefs better than false beliefs. This is not surprising as false beliefs require the ability to understand someone else's behavior, which remains a complex task for participants with DS (Wellman, Cross, & Watson, 2001). Furthermore, false beliefs require pragmatic skills, which develop after the age of 5 in TD children (Westra & Carruthers, 2017). However, it should be noted that two of our subjects, the ones with a higher NVMA (63 months at the beginning of the study), reached the false-belief comprehension level at the end of the training. Once again, further research with more subjects is necessary to confirm the possibility of high-functioning individuals with DS (whose mental age is higher than 5 years old) of reaching a false-belief comprehension level once they have been trained. Lastly, it seems that DS participants who achieve the best result in ToM (hence, false-belief skills), are also those who perform the best in language tests and who demonstrate good language skills [respective Spearman's rho with lexical age, pretest: $r = -0.46$ ($p = 0.43$) and post-test: $r = 0.95$ ($p = 0.01$)]. These results are consistent with those of de Villiers (2005) and Harris (2005). These authors showed that good language proficiency is an advantage to develop ToM and more specifically false-belief comprehension. Indeed, our DS participants with limited language skills had greater difficulty in understanding stories as well as instructions. We observed that their levels of interpretation of the surrounding world seemed to be quite limited. In summary, good language skills seem to benefit the most from ToM training.

Conclusion

The main goal of this study was to determine the possibility of improving ToM skills in individuals with Down syndrome. We chose to adapt a multimodal material that showed its effectiveness among TD children (Gombert et al., 2016). At the end of a 10-week training session, trained participants with DS performed better on ToM tasks than their non-trained peers. However, we must keep in mind that this is simply a pilot study. As our experimental sample was quite small and the training quite short (only 10 weeks), we cannot generalize our results to the whole DS population. It would then be appropriate to reproduce this design with a larger sample of participants. It would therefore be appropriate to conduct a longer training and to explore the long-term maintenance of the training effects. Finally, it would also be interesting to work separately on emotions, beliefs and false beliefs. The chosen tests would then target a single ToM aspect and give more precise results. Nevertheless, despite the different suggestions for improving the experimental design, our results seem to highlight the possibility of some improvement of ToM abilities among DS people.

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Appendix 1 – ToM Inventory tasks

1. *Emotion recognition task – 4 points*: assesses the child's ability to recognize the four basic emotions (glad, sad, fear and angry) by pointing the appropriate faces.
2. *Desire-based emotion task – 1 point*: assesses the child's ability to understand someone's desire and his/her subsequent emotion.
3. *"Seeing leads know" task – 1 point*: assesses the child's ability to attribute knowledge or ignorance to a character by looking at him/her or hearing him/her.
4. *"Line of sight" task – 2 points*: assesses the child's ability to take someone else's point of view and to understand that people may not see the same thing depending on positioning.
5. *Perception-base action task – 1 point*: assesses the child's ability to understand why an object is in a particular place. In other words, the child must understand that knowledge can be achieved through visual perception ("I see that shoes are on a chair so I know where they are") and that knowledge guides behavior (knowing that those shoes are on a chair leads a person to look for the shoes on the chair).
6. *Standard false-belief task – 1 point*: assesses the child's ability to understand that people can have a belief that contradicts reality (a person may look for an object in a specific location based on his/her belief but it is false by the fact that another person has moved the object).
7. *Belief- and reality-based emotion and second order emotion task – 3 points*: assesses the child's ability to understand the points of view of two different characters as well as the emotional consequences of their respective behaviors.
8. *Message-desire discrepant task – 1 point*: assesses the child's ability to infer someone's belief when interpreting desire of a third person in the context of change of location. In other words, is the child able to understand that character A can infer that character B will be disappointed when he/she does not find an object where he/she thought it was?
9. Second-order false-belief task – *1 point*: assesses the child's ability to understand that a person may have another thought than him-/herself and that a third person may also think differently.

Appendix 2 – Training activities

1. *Shared reading*: Two children's books were alternately used with all participants to work on emotions, beliefs and false beliefs. Before running shared reading, we conducted a picture naming task to ensure that words used in the stories were known by the participants. The experimenter reads the book with the participant and asks him/her questions about the story and about the characters' emotions and beliefs
2. *Emotion assignment*: Participants were presented with drawings representing different life situations in which the face of the main character was erased. Participants had to choose among several faces which one matched with the emotion felt by the character.
3. *Symbolic game*: Participants were presented with a set of 10 pictures representing a situation to act out (e.g., flying, cooking, sleeping, dancing, etc.). The experimenter had to guess the situation.
4. *Referential communication task*: As in the Glucksberg's experiment (Glucksberg, Kauss, & Weisberg, 1966), the participant and the experimenter are separated by an opaque screen. They alternatively play the role of a locator or receptor. The speaker must guide the receptor so that he/she can place pictures at a specific place on a board.