

Understanding motor acts and motor intentions in Williams syndrome

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ABSTRACT

Williams syndrome (WS) is a rare genetic disorder associated with unusually hyper-social demeanor and ease with strangers. These personality traits are accompanied by difficulties in social interactions, possibly related, at least in part, to a difficulty in understanding others' mental states. Studies on mentalizing capacities in individuals with WS have often led to contrasting results, some studies revealing specific impairments, others highlighting spared mentalizing capacities. So far, however, no study investigated the performance of individuals with WS in non-inferential understanding of others' motor intentions. In the present study we investigated this capacity by using a computer-based behavioral task using pictures of hand–object interactions. We asked individuals with WS first to describe what the other was doing (i.e. a task implying no kind of intention reading), and secondly, if successful in answering the first question, to describe the motor intention underlying the observed motor acts (i.e. why an act was being done, a task requiring non-inferential motor intention understanding). Results showed that individuals with WS made more errors in understanding what the other was doing (i.e. understanding a motor act) compared to both mental-age matched controls and chronological-age matched peers with typical development, while showing mental-age appropriate performance in understanding why an individual was acting (i.e. understanding a motor intention). These findings suggest novel perspectives for understanding impairments in social behavior in WS.

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1. Introduction

Williams syndrome (WS) is a genetic disorder caused by a hemizygous deletion in chromosome band 7q11.23, including the gene for elastin (ELN) and about 20 surrounding genes (Ewart et al., 1993; Korenberg et al., 2000). Incidence was evaluated as 1:20,000 live births (Morris, Demsey, Leonard, Dilts, & Blackburn, 1988), but more recent estimates speak of 1:7500 live births (Stromme, Bjornstad, & Ramstad, 2002). WS is associated with relatively low IQ (Mervis et al., 2000; Searcy et al., 2004; Vicari, Carlesimo, Brizzolara, & Pezzini, 1996) and a neuropsychological profile of strengths and weaknesses in both verbal and visuo-spatial domains (Bellugi, Lichtenberger, Jones, Lai, & St George, 2001; Brock, 2007; Levy & Bechar, 2003; Vicari, Caselli, Gagliardi, Tonucci, & Volterra, 2002; Volterra, Capirci, Pezzini, Sabbadini, & Vicari, 1996).

A consistent behavioral characteristic of individuals with WS is their hyper-social character (Bellugi, Adolphs, Cassady, & Chiles,

1999; Mervis & Klein-Tasman, 2000). Children with WS are often described as displaying a natural drive toward others (Doyle, Bellugi, Korenberg, & Graham, 2003), being gregarious, people-oriented and sensitive (Klein-Tasman & Mervis, 2003). Nonetheless, this outgoing personality does not result in appropriate behaviors in social contexts, as individuals with WS tend to approach others in an indiscriminate over-friendly way. Some studies ascribe this behavior to an overreliance on evident social signals, typically viewed as positive cues (e.g. a smiling face), and parallel inability to catch more subtle ones (e.g. furrowed eyebrows) (Bellugi et al., 1999; Jones et al., 2000). Other studies suggest that these hyper-social behaviors are due to the presence of a strong drive toward social stimuli and to a lack of inhibition (Frigerio et al., 2006; Mervis et al., 2003). Even if their cause is still unclear, these behaviors lead to lack of social adjustment in comparison to controls with typical development (TD) (Gosh & Pankau, 1997), anxiety (Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis, 2006) and social exploitation (Udwin, Howlin, Davies, & Mannion, 1998). This evidence has led researchers to investigate the mechanisms that may lead to impairment in social cognition in this syndrome (Frigerio et al., 2006; Riby & Hancock, 2008) and to consider that mental states attribution might be an area of weakness for individuals with

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WS (Porter, Coltheart, & Langdon, 2008; Santos & Deruelle, 2009; Sullivan & Tager-Flusberg, 1999; Tager-Flusberg & Sullivan, 2000; Tager-Flusberg, Sullivan, & Boshart, 1997).

Mental states attribution (also termed mentalizing) indicates the ability to make inferences about others' mental states and has been described as involving several brain regions, which allow inferring the goals and intentions of others' overt actions (Frith & Frith, 2006). Mentalizing abilities in our everyday social life, appear to be mediated by various mechanisms. On one hand recent studies have shown that observing others actions elicits a matching activation in the observer's parieto-frontal motor system, allowing for immediate, non-inferential motor intention understanding (see Rizzolatti & Sinigaglia, 2010 for a review). On the other hand this matching motor-activation does not appear to account for more complex mentalizing abilities (Frith & Frith, 2003; Saxe, Carey, & Kanwisher, 2004). For example, when an observed action is unfamiliar or bizarre, other higher-level, top-down structures are involved, among which the rostral cingulate cortex and the region around the superior temporal sulcus (Brass, Schmitt, Spengler, & Geyerly, 2007; Liepelt, Von Cramon, & Brass, 2008). Over the last years, brain-imaging studies have shown how non-inferential motor knowledge and inferential mentalizing mechanisms, may play distinct but complementary roles in understanding goals and intentions of others' actions (de Lange, Spronk, Willems, Toni, & Bekkering, 2008; Hari & Kujala, 2009).

In WS, behavioral studies have highlighted an uneven performance in mental state attribution (Porter et al., 2008; Santos & Deruelle, 2009; Tager-Flusberg & Sullivan, 2000), but we know of no study specifically dedicated to understanding of *motor* intention. The present study aims to better our knowledge of this type of intention understanding in youths with WS relative to TD children, by explicitly analyzing the understanding of observed actions.

The term motor intention has been used by developmental psychologists to indicate understanding a *single motor act* as goal-directed as well as understanding a *series of motor acts* as determined by overarching goals (Gergerly & Csibra, 2003; Gerson & Woodward, 2010; Rochat, 2007; Sommerville, Hildebrand, & Crane, 2008; Sommerville & Woodward, 2005; Sommerville, Woodward, & Needham, 2005; Woodward & Sommerville, 2000; Woodward, Sommerville, Gerson, Henderson, & Buresh, 2009). However a conceptual distinction may be drawn between these two conditions. For example, imagine observing a person grasping a mug. We may perceive the goal-relatedness of each one of his motor acts (e.g. reaching for the mug, grasping it, picking it up, etc.). Furthermore, depending on how the mug is being grasped (e.g. shaping the hand as a hook around its handle or wrapping thumb and four fingers as a claw around its upper edge) and in which context it is being grasped (e.g. close to a teapot or close to a dish drainer), these motor acts may be understood as part of a motor act sequence with an overarching goal (e.g. drinking tea or putting the mug away). In the first case we have an immediate understanding of *what* the other is doing (i.e. reaching, grasping, picking up, etc.), while in the second case we grasp the motor intention underlying the observed motor act, i.e. the *why* of the other's actions (i.e. drinking, putting away, etc.).

Neurophysiological research on the functional properties of the parieto-frontal hand grasping/manipulations circuit sustained the importance of the *what/why* distinction in immediate, non-inferential, understanding of others' actions. In particular, research on the properties of mirror neurons in the parieto-frontal circuit of non-human primates, showed that these neurons encode the observed motor acts in relation to their goals, i.e. they encode *what* the others are doing (e.g. grasping, holding, or breaking) (Gallese, Fadiga, Fogassi, & Rizzolatti, 1996; Rizzolatti, Fadiga, Gallese, & Fogassi, 1996; Umiltà et al., 2008). Subsequent studies uncovered a chained organization in motor acts sequence both during action

execution and observation. Namely, the same goal-related motor acts (e.g. grasping) are encoded by different neurons depending on the over-arching goal of the action sequence in which they are embedded (e.g. grasping-to-eat or grasping-to-place). Thus, according to the context in which they are performed and other variables, the observation of an initial motor act of a sequence activates the whole motor sequence in the parieto-frontal areas of the observers, allowing them to understand, from the very beginning, the motor intention underlying the overarching action, i.e. *why* an action is performed (Fogassi et al., 2005; Iacoboni et al., 2005). Ensuing neuro-imaging and EEG studies have traced similar circuits in humans. These circuits become active at the mere sight of static stimuli showing specific hand-object interactions. Different parts of these circuits encode the *what* and the *why* of an observed action (Iacoboni et al., 2005; Johnson-Frey et al., 2003). As stated above these are not the only circuits involved in mentalizing abilities. For example a recent fMRI study by Spunt and colleagues has shown how other brain areas (i.e. the dorsomedial and the ventromedial pre-frontal cortex, the temporal poles bilaterally and the posterior cingulate cortex) may play a role in understanding the actions of others. These authors highlight how the mirror neuron system encodes immediate, motor aspects of others' intentional actions, while other mentalizing systems interpret these aspects in terms of unobservable mental states and traits (Spunt, Satpute, & Lieberman, 2010).

Following these developmental and neurophysiological studies a recent behavioral study (Boria et al., 2009) has evaluated *what* and *why* understanding in a group of high functioning children with autistic spectrum disorders (HFA) and matched TD controls. The task used static images showing different hand-object interactions to discriminate understanding the goal-relatedness of a single action (i.e. touching vs. grasping) and understanding the motor intention with which it was being performed (i.e. grasping to use vs. grasping to put away). Interestingly, results highlighted a specific impairment in *why* understanding as opposed to *what* understanding in children with HFA, which could be overcome through contextual cueing.

Summing up, increasing evidence from developmental psychology, neurophysiology and research on neuro-developmental disorders, has highlighted the relevance of distinguishing between understanding the *what* and the *why* of an observed act. However, so far no behavioral study has attempted to assess these basic abilities in WS. Therefore we attempted to gain better knowledge of motor acts and motor intention understanding in youths with WS relative to typically developing controls, by focusing on the following questions: (1) Are individuals with WS able to recognize different observed motor acts (i.e. touching or grasping)? (2) Are they able to interpret these acts in terms of overarching goals (i.e. grasping-to-use or grasping-to-place)?

To address these issues we studied the performance of a group of youths with WS and a TD control group, individually matched on mental age and lexical comprehension, on a task inspired by Boria et al. (2009). Further, to fully examine motor intention understanding in WS and to discriminate between impairment and mental-age appropriate performance we also included a group of TD youths individually matched for chronological age and gender.

2. Materials and methods

2.1. Participants

A total of 55 participants were recruited to the study: 19 youths with WS, 18 children with TD individually matched on mental age, lexical comprehension and gender (TDMA) and 18 youths with TD individually matched on chronological age and gender (TDCA). As 1 child with WS was excluded from the study for failing to comply with task requirements during training phase, the final sample comprised: 18 youths with WS (9 females and 9 males with mean chronological age 13;7, range 5;1 to 30;0 years, and mean mental age 6;5, range 4;1 to 11;2), 18 children with

Table 1
Demographics of the participants.

	WS	TDMA	TDCA
N	18	18	18
Gender	9:9 ^a	9:9	9:9
Chronological age	13;7 ± 6.7 ^b	6;1 ± 1.5	13;6 ± 6.7
Leiter-R mental age	6;5 ± 1.9	6;6 ± 1.9	–
Leiter-R IQ	63.3 ± 12.4 ^c	109.8 ± 12.7	–
PPVT-R lexical age	6;9 ± 2.4	6;6 ± 2.5	–
PPVT-R standard score	78.6 ± 10.6	101.6 ± 16.3	–

^a Males:females.

^b Mean years;months ± standard deviation.

^c Mean ± standard deviation.

TDMA (9 females and 9 males with mean chronological age 6;1, range 3;7 to 10;2 years, and mean mental age 6;6, range 4;0 to 11;5) and 18 youths with TDCA (9 females and 9 males with mean chronological age 13;6, range 5;4 to 30;1). Characteristics of the sample are summarized in Table 1. Receptive lexical age and mental age in the WS and TDMA groups were evaluated using respectively: the Peabody Picture Vocabulary Test (PPVT-R) (Dunn & Dunn, 1981; Stella, Pizzoli, & Tressoldi, 2000) and the Leiter International Performance Scale-Revised (Leiter-R) (Roid & Miller, 2002). Brief version of the Leiter-R was used (four out of 10 subtests: Figure Ground, Form Completion, Sequential Order and Repeated Patterns), that yields an IQ brief and allows computing corresponding MA. One-way ANOVA revealed no significant Group differences on MA ($F_{(1,34)} = .012$; $p = .914$, $\eta_p^2 = .000$) and lexical comprehension ($F_{(1,34)} = .158$; $p = .694$, $\eta_p^2 = .005$) between the WS and the TDMA groups. One-way ANOVA showed no significant Group differences on chronological age ($F_{(1,34)} = .002$; $p = .969$, $\eta_p^2 = .000$) between the WS and TDCA groups. Participants with WS attended the Children's Hospital "Bambino Gesù" of Rome for clinical follow-up. All of them exhibited a positive clinical diagnosis; furthermore, all of them had a positive diagnosis established through FISH genetic test for microdeletion of specific gene markers; absence of neurosensory deficits; absence of epilepsy and psychopathological disorders. All of them lived with their own families. TD children were recruited from a preschool in Parma, an elementary school in Rome and word of mouth through research personnel. They were all native speakers of Italian. Experiments were approved by the local ethical committee (Comitato Etico Unico per la Provincia di Parma) and conducted according to the Helsinki declaration. Informed consent was obtained for all participants before taking part in the study.

2.2. Design

All participants performed two experiments, during which they observed some pictures being presented to them on a computer screen and answered some questions. Experiment 1 included three tasks: object recognition task, what task and why task. Experiment 2 comprised two tasks: object recognition task and why task. Both experiments involved verbal instructions given by the experimenter. When questions involved a choice among alternative solutions, terms indicating the alternatives were presented in a random order. All sessions were videotaped for later scoring.

2.3. Stimuli

Object recognition tasks comprised pictures of objects, shown on a neutral background (a total of 15 objects for Experiment 1, 17 objects for Experiment 2). (View Supplementary Materials Tables 5 and 6 for a full list of objects shown respectively in Experiments 1 and 2).

Experiment 1 further comprised a set of 45 pictures, in which each one of the 15 objects present in the object recognition task was shown once in presence of a hand touching the object (15 pictures in total) and once with a hand grasping the object (30 pictures in total), 15 pictures showing grasping to use and 15 pictures showing grasping to put it away. The first experiment was preceded by a training phase, in which 2 real objects (a plastic cup and a fork) and 3 pictured objects (a glass, a fork and a felt-tip pen), were presented, each one in the three different situations mentioned above (i.e. touching, grasping to use and grasping to put away).

Experiment 2 further comprised a set of 34 pictures showing each one of the 17 objects present in the object recognition task either in presence of a hand grasping it within a context that indicated the object's use (17 pictures) or within a context that indicated its being placed away (17 pictures). Also Experiment 2 was preceded by a training phase, in which 3 pictures of different objects (a colored pencil, a doll dress and a toy saw) were shown, each one in the two differing context conditions mentioned above (i.e. context indicating object use and context indicating object being placed away).

The main difference between the stimuli presented in Experiments 1 and 2 was that in Experiment 1 hand shape was manipulated within object type in absence of other contextual cues, in Experiment 2 contextual cues were added and manipulated within object type, while hand shape was kept constant. Samples of hand shape

manipulation for Experiment 1 are shown in Fig. 1 (View Supplementary Materials Tables 3 and 4 for complete stimulus set for Experiments 1 and 2 respectively).

In Experiment 1 pictures showing a hand touching the object could show one of five different hand shapes. All hand shapes are presented in Fig. 1. The five different hand shapes were used so as to avoid practice effects and they were all chosen so as to avoid confounds with grasping of the object.

Pictures showing grasping of the object in Experiment 1 are described following grasping taxonomy constructed by Cutkosky, which takes into account object function and has been used in describing grasping of everyday household items and tools (Cutkosky, 1989; Zheng, De La Rosa, & Dollar, 2011). Accordingly, pictures showing a hand grasping the object to use it could show one of four types of grasps and pictures showing a hand grasping the object to put it away could show one of three types of grasps. All hand shapes are presented in Fig. 1.

High-definition pictures were custom-made using a 4.0 mega pixel Nikon Coolpix 4500 camera; they were modeled after Boria et al. (2009), but modified using common household items which could be familiar for participants' age groups. To test their validity, the pictures were shown, before presenting them to the experimental groups, to a group of 16 TD adult volunteers. The data showed 98% of overall correct answers for the stimuli used in Experiment 1 and 94% for the stimuli used in Experiment 2. Pictures used in the object recognition task were 2.7 in. × 2.3 in. image size; while pictures used in the study phase were cropped to 7.8 in. × 5.9 in. image size using Adobe Photoshop 9.0. All images had a 300 dpi resolution. In all pictures, objects—hand or object—hand-context were placed on a white background surface to increase contrast. During training and study phases a 19 in. computer screen was used and images were shown using Microsoft PowerPoint slides.

2.4. Procedures

Individual testing sessions took place at the Children's Hospital "Bambino Gesù" in Rome for the WS group and in familiar educational environments for the TD groups. Participants were tested in a single session, with the exception of younger children who were allowed for a short break between sessions to avoid excessive fatigue. Individuals in all groups were invited to sit at a table next to the experimenter in front of the computer screen. Study phases for both experiments included 2 orders of presentation of test item conditions to avoid fatigue and practice effects.

2.4.1. Experiment 1

Training phase required the experimenter to present initially an object on the table in clear view of the participant asking her/him to recognize it by saying, "What is this?". Then the experimenter demonstrated 3 different object–hand conditions (i.e. either touching the object, grasping the object to use it or grasping the object to put it away), one at the time, asking in each condition: "What am I doing? Am I touching or grasping?" and, in grasping conditions, further asking, "Why am I grasping it? To use it or to put it away?". Children were allowed to take as much time as they wanted to answer and when questions involved a choice among alternative solutions, terms indicating the alternatives were presented in a random order (e.g. "What is she doing? Touching or grasping?" or "What is she doing? Grasping or touching?").

Once this initial training with real objects was completed an equivalent training with pictures on a computer screen was carried out. The experimenter introduced it by stating: "Now we are going to do the same thing with the computer; I'm going to show you some objects on the screen and a person interacting with them and I'll ask you some questions". The first picture presented on the computer screen during this second part of the training phase showed one of the two real objects seen in the first part, so as to obtain a smooth transition from real life objects to pictures presented on the screen. The participant was shown a picture on the upper left corner of the screen depicting an object and asked to recognize the object (object recognition task). If participants provided an incorrect answer, they were offered correct labeling of the object by the experimenter and were allowed to proceed to the second question. Next a larger picture was shown in the center of the screen displaying the same object and a hand touching, grasping to use or grasping to put away the recognized object (object–hand conditions were presented in a pseudo-random order). While viewing this second image participants were asked: "What is she doing? Touching or grasping?" (what task), and further asked, only in the grasping condition, "Why is she grasping it? To use it or to put it away?" (why task). Terms indicating alternative solutions were presented in a random order in both tasks (e.g. "Why is she grasping it? To use it or to put it away?" or "Why is she grasping it? To put it away or to use it?"). During training phase, if participants provided incorrect responses, appropriate feedback was offered by the experimenter, to ensure that participants understood the fundamentals of the experimental task and to avoid any linguistic confounds between 'touching' and 'grasping'. All participants complied with task requirements during training phase, except for one child with WS who was therefore excluded from the study.

Study phase was carried out in the same way as the training phase with pictures. Therefore a picture of an object was shown on the upper left corner of the screen (for a total of 15 different objects) followed by an image of the same object being either touched, grasped-to-use or grasped-to-put-away (for a total of 45 different images) in pseudo-random order and object recognition task, what task and why task were carried out as described above. The only difference being in the fact that participants had access to the second question (why task) if and only if they responded correctly

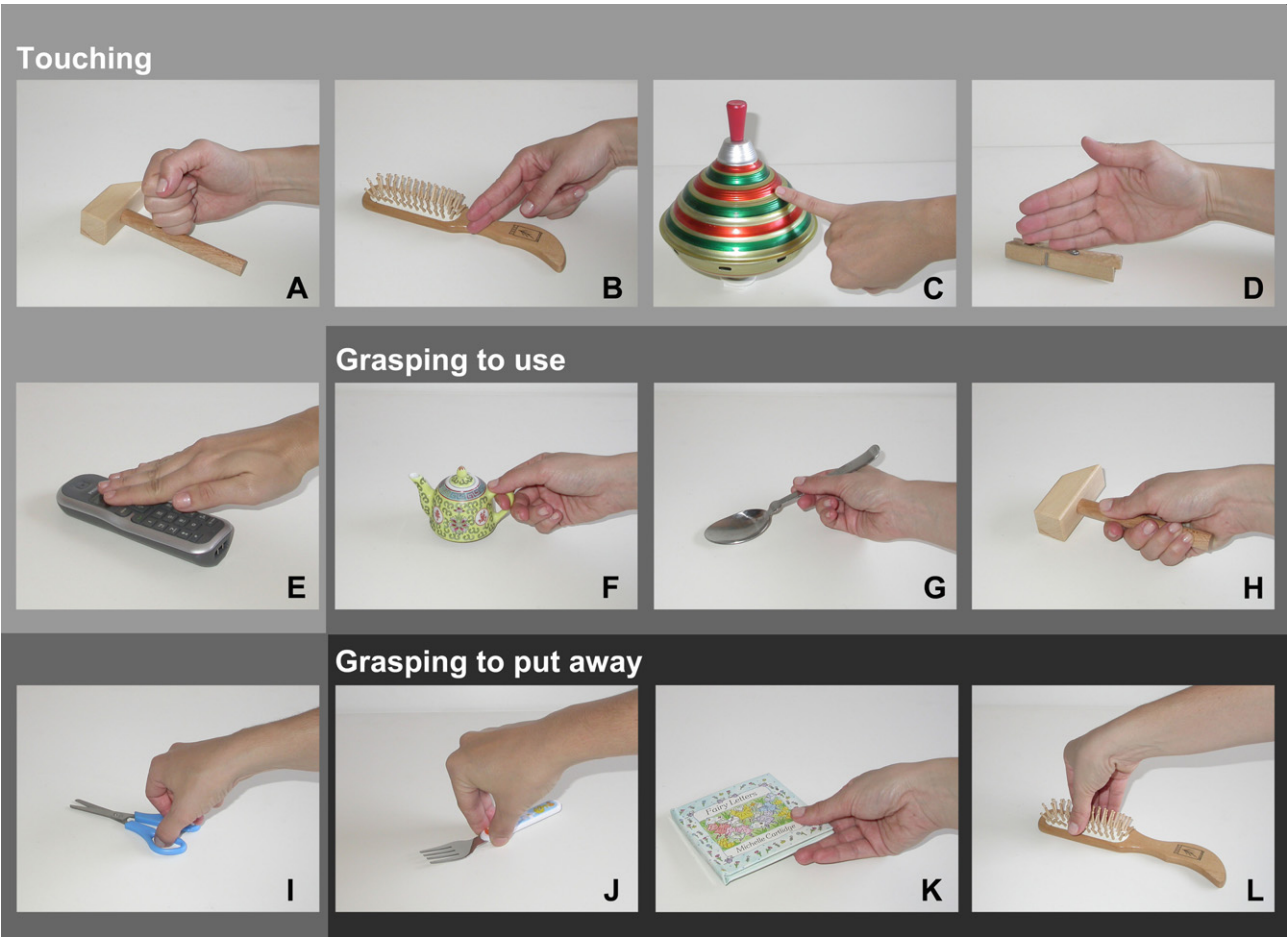


Fig. 1. Hand shapes for Experiment 1. Five images of hand touching the object: (A) closed fist with little finger touching the object, (B) index and middle finger extended other fingers closed with middle finger touching the object, (C) fingers closed with little finger extended and touching the object, (D) all fingers extended together with little finger touching the object; (E) all fingers extended together with index and middle finger touching the object. Four images of hand grasping-to-use the object: (F) hook, (G) light tool wrap, (H) abducted thumb wrap, (I) thumb-index finger grasp. Three images of hand grasping-to-put-away the object: (J) thumb-two finger grasp; (K) thumb-three finger grasp; (L) thumb-four finger grasp.

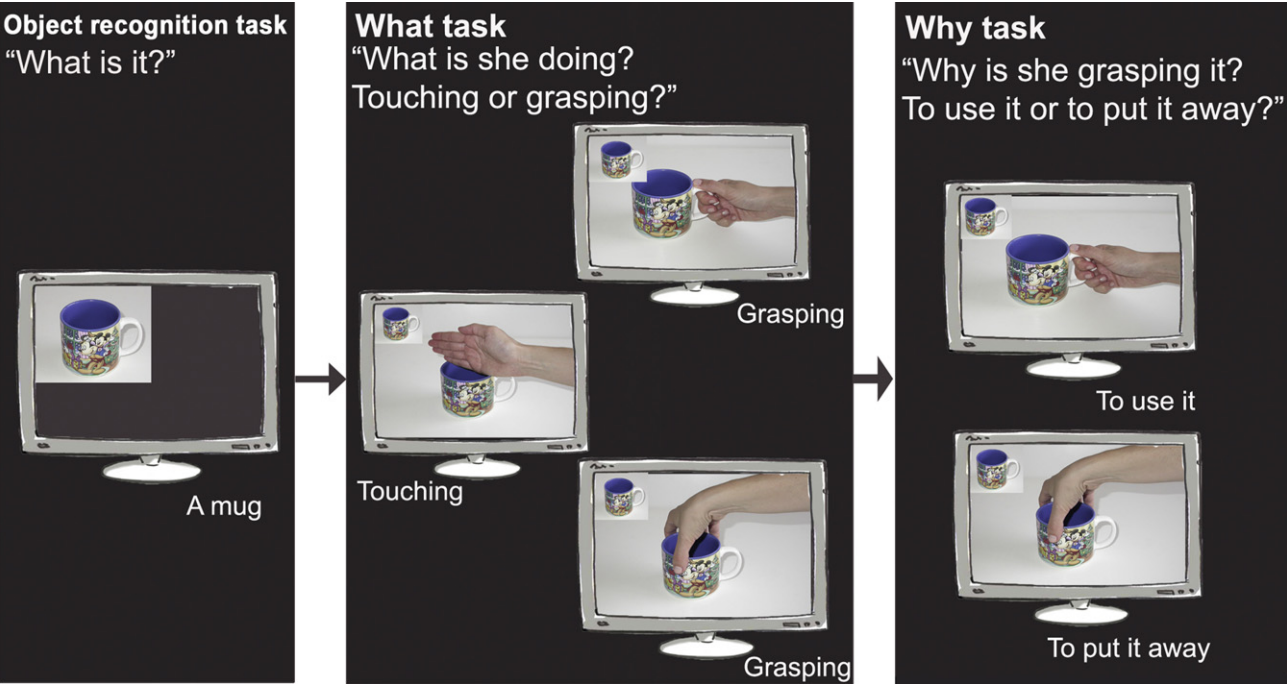


Fig. 2. Experiment 1. Sample study phase.

Table 2
Number of items and error percentage scores tested for each group at each stage for Experiment 1 and 2.

	WS	TDMA	TDCA
Experiment 1			
Object recognition task			
No. of items	15	15	15
Error rate	18.52%	15.56%	5.56%
What task			
Overall no. of items	45	45	45
Overall error rate	17.90%	4.07%	2.72%
Touching no. of items	15	15	15
What-touch error rate	10.37%	1.48%	0.37%
Grasping no. of items	30	30	30
What-grasp error rate	21.67%	5.37%	3.89%
Why task			
Overall mean no. of items	24	28	29
Error rate	33.38%	29.45%	6.44%
Grasp-to-use mean no. of items	12	14	15
Why-use error rate	19.15%	17.60%	6.12%
Grasp-to-place mean no. of items	12	14	14
Why-place error rate	49.77%	40.68%	6.80%
Experiment 2			
Object recognition task			
No. of items	17	17	17
error rate	17.32%	13.40%	5.23%
Why task			
No. of items	34	34	34
Overall error rate	9.80%	9.15%	3.11%

to the first question (what task) in the grasping condition. During both training and study phase children were allowed to take as much time as they wanted to answer. A sample of the study phase for Experiment 1 is illustrated in Fig. 2.

Scoring was based on verbal responses. Reliability between two independent coders (the first and second authors) was assessed for object recognition task and why task. Agreement between coders was 95% for the object recognition task and 98% for the why task. Instances of disagreement were identified and a third coder was requested to code the answers, choosing one of two classifications proposed by the first two coders. The what task was essentially a forced choice alternative, therefore no confounds emerged between coders.

In the object recognition task answers to the first occurrence of an object were categorized as correct or incorrect. Object recognition task was meant to assess participants' overall familiarity with an object, therefore children were considered as having correctly recognized the object if they either provided appropriate labeling for it or offered a correct verbal indication of its proper function (e.g. "book", "fairy-tale" or "to read" when viewing the object 'book'). Responses were considered incorrect if the participant provided inappropriate labeling, improper object function or no response (e.g. "picture frame", "to hang" or "don't know" for the object 'book'). Error percentage was calculated on the total number of different objects viewed (i.e. 15 objects).

In the what task answers were categorized as correct or incorrect. Responses were considered correct if the participant answered "touching" or "grasping" to the appropriate picture; incorrect if the participant answered "grasping" to a picture depicting a touching condition (what-touch errors) and vice versa (what-grasp errors). Error percentage was calculated on the total number pictures shown (i.e. 45 images).

In the why task responses were categorized as correct or incorrect. Answers were considered correct if the participants answered indicating in the observed act the intention canonically related to the object's use (e.g. "to drink" for a mug, "to play" for a spinning top) in response to a grasping-to-use condition, or if she/he answered indicating an intention to put away the object in response to a grasping-to-put-away condition. Responses were considered incorrect if the participant indicated as the other's intention that of using the object while observing a grasping-to-put-away condition (why-use errors) and vice versa (why-place errors). Since participants had access to the why task if and only if they offered a correct answer in the what task in a grasping condition, percentage of why task errors was individually calculated on the number of times that each child had access to the why task. While percentage of why-use errors and why-place errors for each participant was individually calculated on the number of accesses to an image depicting respectively a grasping-to-put-away or a grasping-to-use condition. Number of items and error percentage scores tested for each group at each stage for Experiment 1 are summarized in Table 2.

2.4.2. Experiment 2

Training phase was introduced by the experimenter stating, "I am going to show you some more objects on the screen and a person interacting with them and I'll ask you some questions". A training phase followed, using 3 different objects (i.e.

a toy saw, a corn cob and a doll dress). Participants were initially shown a picture of an object on the upper left corner of the screen and asked to recognize it (object recognition task). If participants provided an incorrect answer, they were offered correct labeling of the object by the experimenter and were allowed to proceed to the second question. Then they were shown a larger picture in the center of the screen showing a hand grasping the object in the same way in one of two differing contexts (i.e. a context indicating object use or a context indicating object being placed away). While viewing this image participants were asked, "Why is she grasping the object, to use it or to put it away?". Alternative solutions were presented in a random order (i.e. "Why is she grasping it? To use it or to put it away?" or "Why is she grasping it? To put it away or to use it?") (why task).

Study phase was analogous to training phase, i.e. participants were shown an object on the upper left corner of the screen and asked to recognize it (for a total of 17 different objects), then the same object was shown in one of the two conditions described above (context indicating object use, context indicating object being placed away) in pseudo-random order (for a total of 34 different images) and for each one object recognition task and why task were carried out as described above. Participants were allowed to take as much time as they wanted to answer during both phases. A sample of the study phase for Experiment 2 is shown in Fig. 3.

Scoring was based on verbal responses. In the object recognition task answers to the first occurrence of an object were categorized as correct or incorrect using the same criteria as in Experiment 1. Error percentage was calculated on the total number of different objects viewed (i.e. 17 objects).

In the why task the participant's response was considered correct if her/his answer was an indication of the agent's intentions as suggested by the context (e.g. "to put away" for a picture showing a hand holding a red toy shirt next to a toy wardrobe). Responses were considered incorrect if participants indicated in the observed act an intention non consistent with the given context (e.g. "to put away" for an image showing a hand holding a red toy shirt next to a Pooh bear missing its shirt). Error percentage was calculated on the total number of images viewed (i.e. 34 pictures). Number of items and error percentage scores tested for each group at each stage for Experiment 1 are summarized in Table 2.

3. Data analysis

All variables of interest were calculated as proportions. An arc-sine transformation was performed on these proportions prior to further analysis in order to meet criteria for parametric analysis; however all percentages reported in both text and figures reflect the untransformed means. All statistical analyses were carried out using the Statistical Package for the Social Sciences (IBM SPSS Inc., Chicago, IL, USA), release 19.0.0. An alpha level of .05 was used to reject the null hypothesis. Effect size (partial eta square η_p^2) was included and effect size lower than .01 was used to consider percentage of variance as negligible (Cohen, 1988). Two one-way ANOVAs were run to investigate the effect of Group (independent variable) on the Percentages of errors in the object recognition tasks (dependent variable) of Experiments 1 and 2. For Experiment 1 repeated measures ANOVA was used to evaluate the effect of object recognition (independent variable) on subsequent what and why tasks performance (dependent variable), while for Experiment 2 one-way ANOVA was used to evaluate the effect of object recognition (independent variable) on subsequent why tasks performance (dependent variable). For Experiment 1 percentages of access to why-use images and why-place images by the two groups of participants were controlled with a one-way ANOVA.

Percentages of errors committed in Experiment 1 were submitted to two-way repeated measures ANOVA with Group (TD and WS) as between-subjects factor and Task (what and why) as within-subjects factor. As follow-up Fisher LSD test were applied. A second repeated measures ANOVA was conducted to explore the difference between Groups (between-subjects factor) on the percentages of errors committed in the why Tasks of Experiments 1 and 2 (within-subjects factor). Repeated measures ANOVA was also used to compare between and within Groups the percentages of error types in the what task (what-grasp and what-touch errors) and why task (why-use and why-place errors) of Experiment 1.

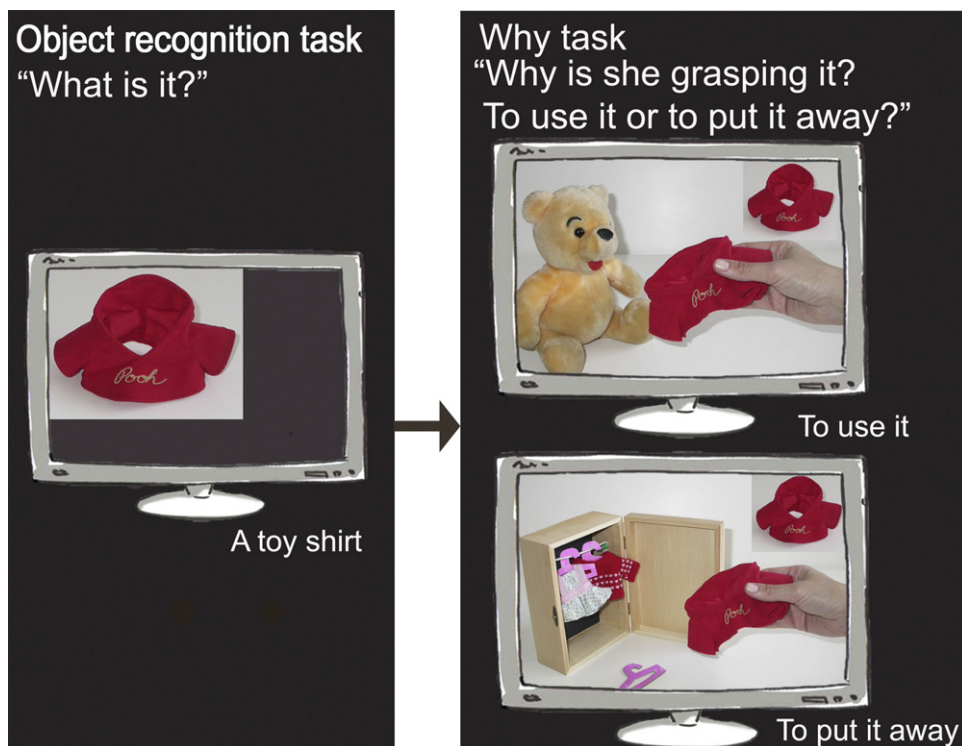


Fig. 3. Experiment 2. Sample study phase.

4. Results

4.1. Experiment 1

4.1.1. Object recognition task

Error rate was 18.52% in the WS group, 15.56% in the TDMA group and 5.56% in the TDCA group. A significant difference emerged among groups (one-way ANOVA, $F_{(2,51)} = 12.941$; $p < .001$, $\eta_p^2 = .337$), post hoc (Fisher LSD test) showed no significant difference between the performance of the WS and TDMA groups ($p = .297$), while the TDCA performed significantly better than the WS and the TDMA groups (both $p < .001$). Group performance in the object recognition task is shown in Fig. 4.

As the object recognition task was meant to assess familiarity with objects, in order to evaluate whether familiarity, or lack thereof, affected subsequent tasks, we conducted a repeated measures ANOVA with object recognition (occurrences of correct and incorrect recognition of the object) as independent variable and

performance in subsequent tasks (error rate in what and why tasks) as dependent variable. Results showed a significant effect of object recognition ($F_{(1,789)} = 28.555$; $p < .001$, $\eta_p^2 = .035$) and error rate in the what and why tasks ($F_{(1,789)} = 325.065$; $p < .001$, $\eta_p^2 = .292$) and a significant interaction between object recognition and error rate in the what and why tasks ($F_{(1,789)} = 11.056$; $p = .001$, $\eta_p^2 = .014$). As post hoc analysis we performed two separate one-way ANOVAs, to evaluate effect size of object recognition on each subsequent task. Both ANOVAs (i.e. object recognition $\times$ error rate in the what task; object recognition $\times$ error rate in the why task), showed a significant effect of object recognition on the what and why tasks respectively ($F_{(1,808)} = 6.173$; $p = .01$, $\eta_p^2 = .008$; $F_{(1,789)} = 25.026$; $p < .001$, $\eta_p^2 = .031$). However, effect size of object recognition in the what task may be considered negligible ($\eta_p^2 < .01$), while it is not negligible in the why task (Cohen, 1988). Effect of incorrect or correct object recognition on performance in subsequent what and why tasks for Experiment 1 is shown in Fig. 5. (View Supplementary Materials Table 5 for mean percentage of errors on individual objects made by each group in the object recognition and on subsequent what and why tasks).

4.1.2. What and why tasks

Mean percentage of errors for the WS group was 17.90% on the what task and 33.38% on the why task, for the TDMA group it was 4.07% on the what task and 29.45% on the why task while for the TDCA group it was 2.72% on the what task and 6.44% on the why task. Repeated measures ANOVA on error rate, with groups (WS, TDMA and TDCA) as between subject factor and task performance (error rate in the what and why tasks) as within-subjects factor, showed an effect of group and task performance (respectively, $F_{(2,51)} = 36.335$; $p < .001$, $\eta_p^2 = .588$; $F_{(1,51)} = 92.388$; $p < .001$, $\eta_p^2 = .644$) and a significant interaction effect, group $\times$ task performance ($F_{(2,51)} = 14.974$; $p < .001$, $\eta_p^2 = .370$). Post hoc analysis (Fisher LSD test) showed that the WS and TDMA groups made significantly more errors in the why task than in the what task ($p < .001$

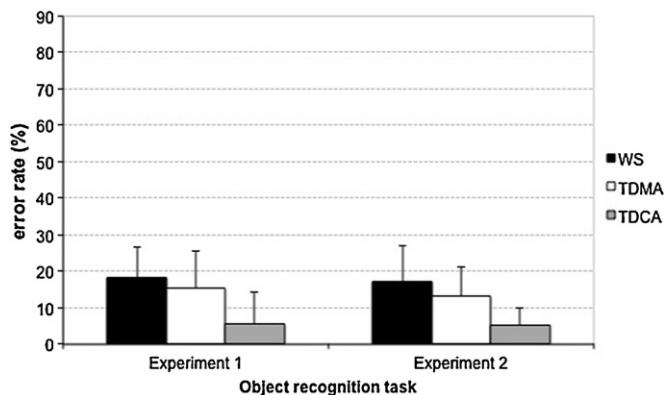


Fig. 4. Error rate in the object recognition task Experiment 1 and 2. Error rates are plotted as percentages for each error type. Error bars indicate standard deviation.

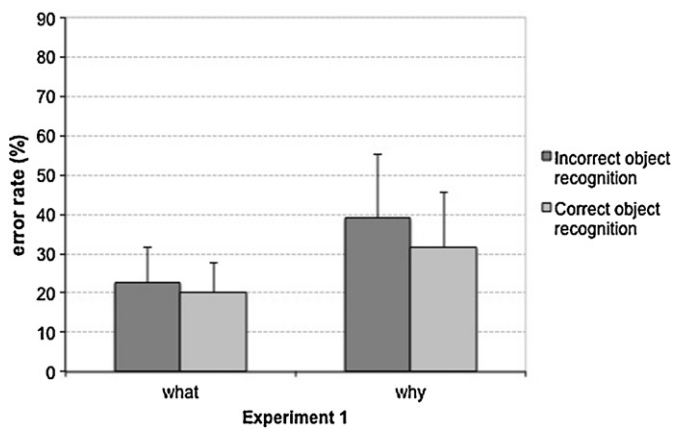


Fig. 5. Effect of incorrect or correct object recognition on performance in subsequent what and why tasks for Experiment 1. Error rates are plotted as percentages for each task type. Error bars indicate standard deviation.

for both groups), while this difference across tasks did not reach significance in the TDCA group ($p = .053$).

Most interestingly, in the what task the WS group differed from both the TDMA and the TDCA groups (both $ps < .001$), while the TDMA and the TDCA did not differ among themselves ($p = .655$). Furthermore, in the why task the WS group differed only from TDCA group ($p < .001$), but not from the TDMA ($p = .368$), while the TDMA and the TDCA groups differed among themselves ($p < .001$).

Percentage of access to why-use images was 52.35% for the WS, 50.77% for the TDMA group and 51.02% for the TDCA group, while percentage of access to why-place images was 47.65% for the WS group, 49.23% for the TDMA group and 48.98% for the TDCA group. Since participants had access to the second question (why task) if and only if they responded correctly to the first question (what task) in the grasping condition, we controlled that percentage of times that children saw why-use images and why-place images did not differ (one-way ANOVA $F_{(2,51)} = .856$; $p = .431$, $\eta_p^2 = .032$).

In the what task participants could make two types of errors: incorrectly responding “touching” when viewing a hand grasping the object (what-grasp errors) and erroneously stating “grasping” upon viewing a hand touching the object (what-touch errors). The amount of what-grasp errors was 21.67% for the WS group, 5.37% for the TDMA group and 3.89% for the TDCA group. The amount of what-touch errors was 10.37% for the WS group, 1.48% for the TDMA group and 0.37% for the TDCA group (see also Table 2).

Repeated measures ANOVA on error rate with group (WS, TDMA and TDCA) as between subject factor and error type (what-grasp and what-touch errors) as within subject factor showed a significant effect of group ($F_{(2,51)} = 20.257$; $p < .001$, $\eta_p^2 = .443$) and error type ($F_{(1,51)} = 8.188$; $p < .01$, $\eta_p^2 = .138$) with more what-grasp errors being made, but no interaction effect between Error type and Group ($F_{(2,51)} = 1.925$; $p = .156$, $\eta_p^2 = .070$). Post hoc showed that the WS group made significantly more errors than both other groups (both $ps < .001$) and no differences emerged between the TDMA and TDCA groups. All groups made more what-grasp errors than what-touch errors (all $ps < .001$).

In the why task participants could make two types of errors: incorrectly responding, “to put away” when viewing a hand grasping the object to use it (why-use errors) and erroneously answering “to use” upon seeing a hand grasping the object to put it away (why-place errors). The amount of why-use errors was 19.15% for the WS group, 17.60% for the TDMA group and 6.12% for the TDCA group. The amount of why-place errors was 49.77% for the WS group, 40.68% for the TDMA group and 6.80% for the TDCA group (see also Table 2).

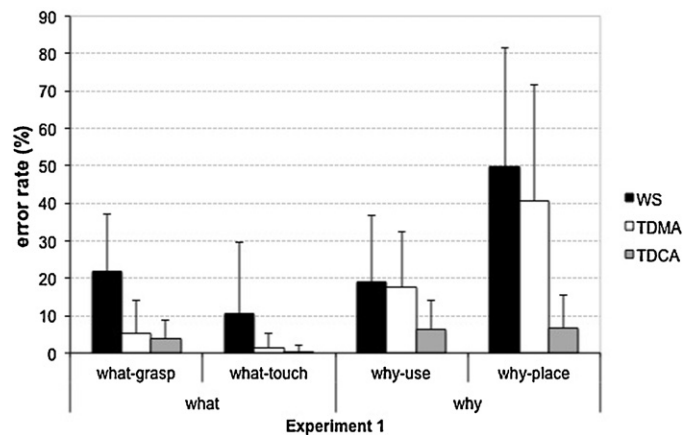


Fig. 6. Error rate in the what and why tasks for Experiment 1. Error rates are plotted as percentages for each error type. Error bars indicate standard deviation.

Repeated measures ANOVA on error rate with Group (WS, TDMA and TDCA) as between subject factor and Error type (why-use errors and why-place errors) as within-subjects factor showed a main effect group ($F_{(2,51)} = 31.069$; $p < .001$, $\eta_p^2 = .549$) and of error type ($F_{(1,51)} = 13.790$; $p = .001$, $\eta_p^2 = .213$) with significantly more why-place errors being made, and significant interaction effect ($F_{(2,51)} = 3.227$; $p = .048$, $\eta_p^2 = .112$). Post hoc (Fisher LSD test) showed that participants in the WS and TDMA group made significantly more why-place errors ($p < .001$) and more why-use errors ($p = .010$ and $p = .019$) than the TDCA group. Furthermore while within the TDCA group the same amount of why-use and why-place errors were made ($p = .907$), both within the WS and the TDMA groups more why-place errors were present ($p < .001$ and $p = .007$ respectively). Mean error rate in the what and why tasks in Experiment 1 for each group is shown in Fig. 6. (View Supplementary Materials Table 5 for mean percentage of errors on individual objects in what and why tasks for each group).

4.2. Experiment 2

4.2.1. Object recognition task

Error rate was 17.32% in the WS group, 13.40% in the TDMA group and 5.23% in the TDCA group. A significant difference emerged between groups (one-way ANOVA, $F_{(2,51)} = 14.103$; $p < .001$, $\eta_p^2 = .356$), post hoc showed no significant difference between the performance of the WS and TDMA groups ($p = .155$), while the TDCA performed significantly better than both WS and TDMA groups ($p < .001$ and $p = .001$ respectively). Group performance in the object recognition task is shown in Fig. 4.

In order to evaluate the effect of familiarity with objects on subsequent why task, we conducted one-way ANOVA on error rate, with object recognition (occurrences of correct and incorrect recognition of the object) as independent variable and why task performance (error rate in the why task) as dependent variable. Results showed a significant effect of object recognition on error rate in the why task ($F_{(1,916)} = 7.078$; $p = .008$, $\eta_p^2 = .008$). However, effect size of object recognition in the why task for Experiment 2 may be considered negligible ($\eta_p^2 < .01$) (Cohen, 1988) (View Supplementary Materials Table 6 for mean percentage of errors on individual objects made by each group in the object recognition task).

4.2.2. Why task

Percentage of incorrect answers was 9.80% for the WS group, 9.15% for the TDMA group and 3.11% for the TDCA group. Repeated measures ANOVA was computed to compare the three groups

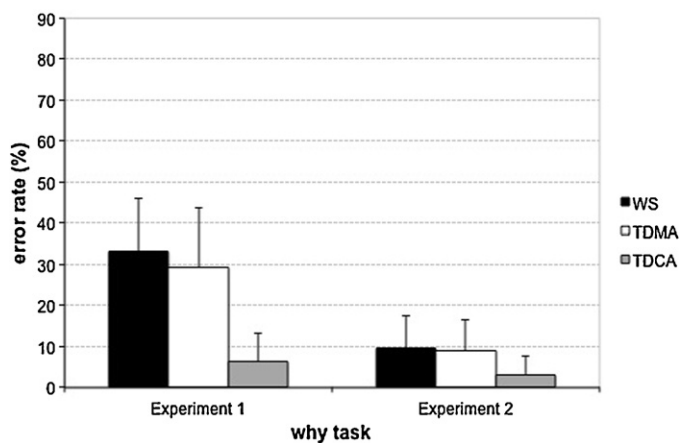


Fig. 7. Error rate in the why tasks for Experiments 1 and 2. Error rates are plotted as percentages for each Experiment. Error bars indicate standard deviation.

(between-subjects factor) on the percentage of errors committed in the why tasks (within-subjects factor) of both experiments (i.e. with or without contextual clues). Results showed a significant effect of groups ($F_{(2,51)} = 27.931$; $p < .001$, $\eta_p^2 = .523$) and tasks ($F_{(1,51)} = 108.445$; $p < .001$, $\eta_p^2 = .680$), as well as significant interaction ($F_{(2,51)} = 12.935$; $p < .001$, $\eta_p^2 = .337$). Post hoc analysis showed performance of the WS and TDMA groups differed from performance of the TDCA group ($p = .015$ and $p = .025$ respectively). Most interestingly both the WS and the TDMA groups were significantly aided by the presence of contextual cues ($p < .001$), while the presence of contextual cueing brought no variation in performance within the TDCA group ($p = .063$). Performance in the why task for Experiments 1 and 2 is shown in Fig. 7 (View Supplementary Materials Table 6 for mean percentage of errors on individual objects made by each group in the why task).

5. Discussion

Despite their hyper-social character and natural drive toward others, individuals with WS often experience difficulties in social interactions (see Martens, Wilson, & Reuters, 2008 for a review). Are these difficulties consequences of specific deficits in mentalizing? In attempting to shed some light on this problem, various studies have examined mentalizing abilities in individuals with WS. The results have been contradictory. While some studies found that in WS mentalizing abilities are above general intellectual capacities (Karmiloff-Smith, Klima, Bellugi, Grant, & Baron-Cohen, 1995; Tager-Flusberg et al., 1997), others report specific impairments (Tager-Flusberg & Sullivan, 2000) and others still highlight mentalizing abilities appropriate to intellectual capacities (Santos & Deruelle, 2009; Sullivan & Tager-Flusberg, 1999). A recent study has stressed heterogeneity among subgroups of individuals with WS (Porter & Coltheart, 2005), maintaining that different mentalizing abilities may correspond to differing profiles (Porter et al., 2008).

In the present study we addressed this issue from a more basic perspective. Rather than examining complex, inferential mentalizing abilities, as done by previous studies, we investigated basic, non-inferential motor intention understanding (see Rizzolatti & Sinigaglia, 2010 for a review). To this purpose we presented individuals with WS with pictures of hand-object interactions and asked them to describe: *what* the other was doing (i.e. what task, not requiring any intention reading), and subsequently, the motor intention underlying the observed motor acts (i.e. why task, requiring motor intention reading).

Results revealed that, in absence of contextual cues (Experiment 1), individuals with WS made a larger amount of errors in the what

task relative to both mental-age and chronological-age matched controls (18% for the WS group vs. 4% for the TDMA group and 3% for the TDCA group), therefore highlighting impaired understanding of *what* the other was doing (e.g. touching or grasping). In the why task percentage of errors for the WS group was higher (33%), yet in the same range as the one shown by mental-age matched TD controls (30%) while differing from chronological-age matched TD controls (6%), therefore showing mental-age appropriate performance. The presence of contextual cues (Experiment 2) markedly decreased errors in the why task for both the WS group and the TDCA controls (error rate dropping to less than 10%), showing that contextual cueing (Experiment 2) is of great aid for these groups.

5.1. Object recognition task

It must be stressed that individuals with WS perfectly understood task requirements and displayed good linguistic understanding of the difference between 'touch' and 'grasp', as demonstrated by their comprehension during training phase, where only one child failed to comply with task requirements. However, we acknowledged a high percentage of errors in the object recognition task and, in order to better understand these error rates, we examined error types in the object recognition task of Experiments 1 and 2. Total errors made in the object recognition task for both experiments included inappropriate labeling, stating improper object function, or no response. Inappropriate labeling and improper object function errors in both experiments were all due to either semantically related errors (i.e. the child offers an incorrect label which belongs to another item semantically related to the observed one, e.g. "toothpaste" for the object 'toothbrush', "comb" for the object 'hairbrush') or visuo-perceptive errors (e.g. the child offers an incorrect label which belongs to another item similar in form or shape to the observed one, e.g. "drill" for the object 'hair drier', "candy" for the object 'bead', "chisel" for the object 'screwdriver'). Both error rates and types of errors were in agreement with those reported in a previous study by Bello and colleagues, which considered performance of a group of children with WS and their mental-age and chronological-age matched controls on the Boston Naming Task (Bello, Capirci, & Volterra, 2004).

Error rates in the object recognition task did not affect the study's main result, given that the WS group displayed an error pattern no different to that of the TDMA group and that all participants were provided with appropriate labeling, by the experimenter, in all cases of a lack of object recognition.

All objects used were common household items well known to children. However the amount of familiarity with a given object, proved to vary to a certain degree among children, with some objects proving to be less familiar than others (as shown in Tables 5 and 6 in Supplementary Materials). We analyzed whether performance on subsequent what and why tasks was effected by this familiarity with the presented object. Results showed that in Experiment 1 the effect size of object recognition on performance in the what task may be considered negligible (effect size was lower than .01), while the effect of object recognition on performance in the why task was more than three times greater.

Therefore the presence of objects, which proved to be less familiar, has allowed us to observe another difference between *what/why* understanding. In fact, the above result may be explained by considering a difference between the two tasks present in Experiment 1. Consider observing and distinguishing between two goal-related motor acts (e.g. touching and grasping) as in the what task, in this case object familiarity is irrelevant. In fact, the information that we need is conveyed by the motor act in itself, i.e. the relation between an object's general geometry, its affordances and the motor act's requirements. For example, a fundamental requisite of the act of prehension is stability, i.e. in order to grasp an object we must

hold it securely (Cutkosky, 1989; Napier, 1956). Therefore, since the motor act of grasping is characterized by stability, while the act of touching merely requires contact between hand and object, decoding the presence of the former or of the latter, will have little to do with how familiar we are with the object. To the contrary, understanding of *why* the object is being grasped, is largely related to how an object is grasped. The observed grip foreshadows a specific object's use and will therefore is largely influenced by how familiar we are with the object. For example, the grasp used to pick up a pencil is entirely different from the one we use to write, but if we possess no familiarity whatsoever with a pencil or with its use, we may not distinguish between the two grasps as they may hardly be understood as part of a motor sequence with an overarching goal.

In relation to Experiment 2, we conducted a similar analysis and results showed that the effect size of object recognition on performance in the *why* task in presence of contextual cues is negligible (effect size was lower than .01) (Cohen, 1988). This result may be due to the fact that contextual cues have an overall aiding effect on distinguishing two grasps in relation to their overarching goals.

5.2. What task

The finding of impairment in the *what* task for WS was rather unexpected. This finding, however, is much less surprising when considering the literature indicating dysfunction of parieto-frontal cortical circuits responsible for visuo-motor transformations in WS (see below). If the data are correct, the difficulty in recognizing motor acts may be accounted for by a poor development of the mechanism dedicated to matching observed motor acts on motor engrams in the inferior parietal lobule and ventral premotor cortex (the mirror mechanism), due to abnormalities of these areas. In fact both clinical and brain-imaging studies on motor impairments in WS appear to support this hypothesis.

There is agreement that the clinical features of WS include developmental delay in motor skills (Lenhoff, Wang, Greenberg, & Bellugi, 1997; Semell & Rosner, 2003; Vicari et al., 2004). Particularly important are some recent data by Stefanini, Bello, Volterra, and Carlier (2008), analyzing fine hand motor actions in children with WS, considering three types of grips: a precision grip (i.e. object prehension with the tips of the index finger and thumb), a lateral grip (i.e. object prehension with the opposition of the thumb to the radial side of the index finger) and a finger grip (i.e. prehension with all fingers). Results showed that individuals with WS frequently used the lateral grip to grasp objects, instead of a precision grip, a behavior in contrast with the one observed in chronological-age matched TD controls and only rarely present in mental-age matched TD controls. The lateral grip is a form of primitive prehension, usually preceding the occurrence of more accurate precision grip. However, the rare occurrence of this grip type in the mental-age matched controls led authors to suggest an atypical behavior in the WS group rather than a delay.

Behavioral data showing impairment in motor functions in WS has been corroborated by neuro-anatomical studies. In particular, an fMRI study by Meyer-Lindenberg et al. (2004), highlighted hypoactivation in the region of the intraparietal sulcus in adults with WS compared to TD controls. This study attempted to rule out confounds met in other fMRI studies due to cognitive delay, by considering a group of adults with genetically confirmed WS, but spared general intelligence, and controls matched for age, gender, handedness and IQ. Consistent with these data, are also findings from structural brain imaging studies that showed abnormalities, in WS individuals, in the fronto-parietal regions involved in the visual guidance of movement (see Hocking, Bradshaw, & Rinehart, 2008). Reduced gray matter volume in both parietal and occipital areas in individuals with WS compared to controls has also been reported (Boddaert et al., 2006; Eckert et al., 2005).

In order to better explore impairment in *what* understanding (i.e. understanding of motor acts) in the WS group, we analyzed error types in the *what* task, i.e. *what-grasp* vs. *what-touch* errors. Results showed that all groups made more errors on 'grasp' items, indicating that impairment in *what* understanding in WS is not due to the presence of a deviant strategy in solving the task, as error types are analogous to the ones made by both mental-age and chronological-age matched children.

5.3. Why task

Results in the *why* task showed that individuals with WS did not differ in the amount of errors from mental-age matched controls, while differing from their chronological-age matched peers. This does not indicate spared understanding of *why* an act is being done in WS, but rather mental-age appropriate performance. It is difficult to provide a final answer to the question whether the difference in *why* understanding between the WS group and their chronological age peers, is due to impaired development of specific mechanisms underlying direct action understanding or, rather, to more general cognitive deficits. Yet, considering the above-mentioned functional and anatomical alterations of the parieto-frontal circuit found in WS, it seems reasonable to admit that dysfunctions of these circuits should play a certain role.

Interpreting this result, shown by WS individuals in the *why* task, may also require considering the development of *why* understanding in typical development. A recent study by Bello et al. (under review) has analyzed *what/why* understanding in TD children from 3 to 7 years of age. Results showed a progressive decrease of errors in the *why* task with age growth, with a significant difference in performance by the age of 6–7 years of age, older children performing significantly better (i.e. 80% of correct answers in the *why* task). Similar results were reported for the TD control group considered in the comparable study by Boria et al. (2009), who found that 8-year-old TD children gave about 85% of correct answers in the *why* task. It is clear therefore that in typical development *why* understanding follows a developmental pattern. Furthermore, these studies indicate that it is important to consider error types in relation to the *why* task. In fact, in typical development the amount of *why-use* errors shows little variation, while the amount of *why-place* errors diminishes with age growth (Bello et al., under review). This indicates that typically developing children initially tend to link objects to their functionally-related actions, but with age growth they become gradually able to use information conveyed by hand grip, relying less on this link. A viable hypothesis is that for younger children objects are strongly linked to the action sequence which complies with their main function (e.g. a fork is grasped to eat), but as they further exploit the environment and become more familiar with the range of actions that may be performed with an object, this link is gradually attenuated (e.g. a fork may be grasped to eat, to feed a doll, etc.).

Results from our mental-age and chronological age matched controls confirm previous findings on typical development, in relation to overall errors as well as in relation to error types. In fact, for the TDCA group the *why* task was not harder to solve than the *what* task as for the other groups. Also, youths in the TDCA group performed significantly better than the younger TDMA group in the *why* task and, most importantly, proved to be the only group within which the amount of *why-use* errors and *why-place* errors did not differ.

In examining the different performance of the WS group with respect to the TDCA group in the *why* task, it is important to note that individuals with WS not only did not differ from the younger mental-age matched controls in the amount of overall errors, but also that they similarly made more *why-place* errors. These data

may indicate in WS, a similar strategy in solving the why task to the one displayed by younger children, i.e. a tendency to link objects to their functionally-related actions.

5.4. Possible comparisons with autism

The present study also suggests that performance of individuals with WS and of individuals with autism in *what* and *why* understanding may follow opposite directions. In fact, while our WS participants showed impairment in *what* understanding (i.e. in motor act understanding) and mental-age appropriate performance in *why* understanding, children with HFA considered in Boria et al. (2009) showed spared *what* understanding and impaired *why* understanding. The literature comparing autism and WS has underscored and examined the presence of overlapping socio-cognitive impairments in WS and autism (Klein-Tasman, Mervis, Lord, & Phillips, 2007; Klein-Tasman, Phillips, Lord, Mervis, & Gallo, 2009; Lincoln, Searcy, Jones, & Lord, 2007; Tager-Flusberg, Plesa Skwerer, & Joseph, 2006), often proving that a similar social outcome may be due to divergent traits (Riby & Hancock, 2008; Rodgers, Riby, Janes, Connolly, McConachie, 2011). But, lacking a direct cross-syndrome comparison, for the time being our considerations may only be speculative and we are only able to suggest the possibility of a divergent pattern in *what/why* understanding, which must be further investigated.

6. Conclusion

In conclusion, the *what/why* distinction has led us to provide some preliminary data on motor acts and motor intention understanding in WS. In relation to *why* understanding youths with WS show no specific delay beyond their mental and lexical age, as they do not differ from the mental-age matched controls. This result indicates that youths with WS show, in grasping the overarching goal of an observed action, a behavior similar to the one present in comparatively younger children. On the other hand, *what* understanding has been found to be significantly impaired in youths with WS in comparison to both mental-age and chronological age matched peers. This result indicates that in youths with WS there is an impairment not only in the execution of motor acts, as shown by previous works, but also in their recognition, suggesting the existence of a link between these capacities. In conclusion, our findings highlight different levels of difficulty in understanding motor acts and motor intentions in WS, providing, initial data on basic, non-inferential abilities, which play a role in social interaction.

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Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at doi:10.1016/j.neuropsychologia.2012.03.019.

References

- Bello, A., Capirci, O., & Volterra, V. (2004). Lexical production in children with Williams syndrome: Spontaneous use of gesture in a naming task. *Neuropsychologia*, 42, 201–213.
- Bello, A., Sparaci, L., Stefanini, S., Boria, S., Volterra, V., & Rizzolatti, G. (under review). Motor intention from the start: What and why understanding in a developmental perspective. *Developmental Psychology*.
- Bellugi, U., Adolphs, R., Cassady, C., & Chiles, M. (1999). Towards the neural basis for hypersociability in a genetic syndrome. *NeuroReport*, 10, 1653–1657.
- Bellugi, U., Lichtenberger, L., Jones, W., Lai, Z., & St George, M. (2001). The neurocognitive profile of Williams syndrome: A complex pattern of strengths and weaknesses. In U. Bellugi, & M. St. George (Eds.), *Journey from cognition to brain to gene* (pp. 1–42). Cambridge, MA: MIT Press.
- Boddaert, N., Mochel, F., Meresse, I., Seidenwurm, D., Cachia, A., Brunelle, F., et al. (2006). Parieto-occipital gray matter abnormalities in children with Williams syndrome. *NeuroImage*, 30, 721–725.
- Boria, S., Fabbri-Destro, M., Cattaneo, L., Sparaci, L., Sinigaglia, C., Cossu, G., et al. (2009). Intention understanding in autism. *Plos One*, 4, e5596.
- Brass, M., Schmitt, R. M., Spengler, S., & Gergely, G. (2007). Investigating action understanding: Inferential processes versus action simulation. *Current Biology*, 17, 2117–2121.
- Brock, J. (2007). Language abilities in Williams syndrome: A critical review. *Development and Psychopathology*, 19, 97–127.
- Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). Hillsdale, NJ: Lawrence Erlbaum.
- Cutkosky, M. R. (1989). On grasp choice, grasp models, and the design of hands for manufacturing tasks. *IEEE Transactions on Robotics and Automation*, 5, 269–279.
- de Lange, F. P., Spronk, M., Willems, R. M., Toni, I., & Bekkering, H. (2008). Complementary systems for understanding action intentions. *Current Biology*, 18, 454–457.
- Doyle, T. F., Bellugi, U., Korenberg, J. R., & Graham, J. (2003). “Everybody in the world is my friend” hypersociability in young children with Williams syndrome. *American Journal of Medical Genetics A*, 124A(3), 263–273.
- Dunn, L. M., & Dunn, L. (1981). *Peabody Picture Vocabulary Test Revised (PPVT-R)*. Circle Pines: American Guidance Service.
- Eckert, M. A., Hu, D., Eliez, S., Bellugi, U., Galaburda, A., Korenberg, J., et al. (2005). Evidence for superior parietal impairment in Williams syndrome. *Neurology*, 64, 152–153.
- Ewart, A. K., Morris, C. A., Atkinson, D., Jin, W., Sternes, K., Spallone, P., et al. (1993). Hemizygosity and the elastin locus in a developmental disorder, Williams syndrome. *Nature Genetics*, 5, 11–16.
- Fogassi, L., Ferrari, P. F., Gesierich, B., Rozzi, S., Chersi, F., & Rizzolatti, G. (2005). Parietal lobe: From action organization to intention understanding. *Science*, 308, 662–667.
- Frigerio, E., Burt, M. D., Gagliardi, C., Cioffi, G., Martelli, S., Perrett, D. I., et al. (2006). Is everybody always my friend? Perception of approachability in Williams syndrome. *Neuropsychologia*, 44, 254–259.
- Frith, C. D., & Frith, U. (2006). The neural basis of mentalizing. *Neuron*, 50, 531–534.
- Frith, U., & Frith, C. D. (2003). Development and neurophysiology of mentalizing. *Philosophical Transactions of the Royal Society London B Biological Sciences*, 358, 459–473.
- Gallese, V., Fadiga, L., Fogassi, L., & Rizzolatti, G. (1996). Action recognition in the premotor cortex. *Brain*, 119, 593–609.
- Gergely, G., & Csibra, G. (2003). Teleological reasoning in infancy: The naïve theory of rational action. *Trends in Cognitive Sciences*, 7, 287–292.
- Gerson, S., & Woodward, A. L. (2010). Building intentional action knowledge with one’s hands. In S. P. Johnson (Ed.), *Neo-constructivism*. Oxford: University Press.
- Gosh, A., & Pankau, R. (1997). Personality characteristics and behavior problems in individuals of different ages with Williams syndrome. *Developmental Medicine and Child Neurology*, 39, 527–533.
- Hari, R., & Kujala, M. V. (2009). Brain basis of human social interaction: From concepts to brain imaging. *Physiological Reviews*, 80, 453–479.
- Hocking, D. R., Bradshaw, J. L., & Rinehart, N. J. (2008). Fronto-parietal and cerebellar contributions to motor dysfunction in Williams syndrome. A review and future directions. *Neuroscience and Behavioral Reviews*, 32, 297–307.
- Iacoboni, M., Molnar-Szakacs, I., Gallese, V., Buccino, G., Mazziotta, J. C., & Rizzolatti, G. (2005). Grasping the intentions of others with one’s own mirror neuron system. *Plos Biology*, 3, e79.
- Johnson-Frey, S. H., Maloof, F. R., Newman-Norlund, R., Farrer, C., Inati, S., & Grafton, S. T. (2003). Actions of hand-object interactions? Human inferior frontal cortex and action observation. *Neuron*, 39, 1053–1058.
- Jones, W., Bellugi, U., Lai, Z., Chiles, M., Reilly, J., Lincoln, A., et al. (2000). Hypersociability in Williams syndrome. *Journal of Cognitive Neuroscience*, 12, 30–46.
- Karmiloff-Smith, A., Klima, E., Bellugi, U., Grant, J., & Baron-Cohen, S. (1995). Is there a social module? Language, face processing, and theory of mind in individuals with Williams syndrome. *Journal of Cognitive Neuroscience*, 7, 196L 208.

- Klein-Tasman, B. P., & Mervis, C. B. (2003). Distinctive personality characteristics of 8-, 9-, and 10-year-olds with Williams syndrome. *Developmental Neuropsychology*, 23, 269–290.
- Klein-Tasman, B. P., Mervis, C. B., Lord, C., & Phillips, K. D. (2007). Socio-communicative deficits in young children with Williams syndrome: Performance on the autism diagnostic observation schedule. *Child Neuropsychology*, 13, 444–467.
- Klein-Tasman, B. P., Phillips, K. D., Lord, C., Mervis, C. B., & Gallo, F. J. (2009). Overlap with the autism spectrum in young children with Williams syndrome. *Journal of Developmental and Behavioral Pediatrics*, 30, 289–299.
- Korenberg, J. R., Chen, X.-N., Hirota, H., Lai, Z., Bellugi, U., Burian, D., et al. (2000). Genome structure and cognitive map of Williams syndrome. *Journal of Cognitive Neuroscience*, 12, 89L 107 (Special Issue).
- Lenhoff, H. M., Wang, P. P., Greenberg, F., & Bellugi, U. (1997). Williams syndrome and the brain. *Scientific American*, 277, 68L 73.
- Levy, Y., & Bechar, T. (2003). Cognitive, lexical and morpho-syntactic profiles of Israeli children with Williams syndrome. *Cortex*, 39, 255L 271.
- Leyfer, O. T., Woodruff-Borden, J., Klein-Tasman, B. J., Fricke, J. S., & Mervis, C. B. (2006). Prevalence of psychiatric disorders in 4 to 16-year-olds with Williams syndrome. *American Journal of Medical Genetics, B*, 141B, 615–622.
- Liepert, R., Von Cramon, D. Y., & Brass, M. (2008). How do we infer others' goals from non-stereotypic actions? The outcome of context-sensitive inferential processing in right inferior parietal and posterior temporal cortex. *Neuroimage*, 43, 784L 792.
- Lincoln, A. J., Searcy, Y. M., Jones, W., & Lord, C. (2007). Social interaction behaviors discriminate young children with autism and Williams syndrome. *Journal of the American Academy of Child and Adolescent Psychiatry*, 46, 323–331.
- Martens, M. A., Wilson, S. J., & Reutens, C. (2008). Williams syndrome: A critical review of the cognitive, behavioral, and neuroanatomical phenotype. *Journal of Child Psychology and Psychiatry*, 49, 576–608.
- Mervis, C. B., & Klein-Tasman, B. P. (2000). Williams syndrome: Cognition, personality, and adaptive behavior. *Mental Retardation and Developmental Disabilities Research Reviews*, 6, 148L 158.
- Mervis, C. B., Morris, C. A., Klein-Tasman, B. P., Bertrand, J., Kwitny, S., Appelbaum, L. G., et al. (2003). Attention characteristics of infants and toddlers with Williams syndrome during triadic interactions. *Developmental Neuropsychology*, 23, 243–268.
- Mervis, C. B., Robinson, B. F., Bertrand, J., Morris, C. A., Klein-Tasman, B. P., & Armstrong, S. C. (2000). The Williams syndrome cognitive profile. *Brain and Cognition*, 44, 604L 628.
- Meyer-Lindenberg, A., Kohn, P., Mervis, C. B., Kippenhan, J. S., Olsen, R. K., Morris, C. A., et al. (2004). Neural basis of genetically determined visuospatial construction deficit in Williams syndrome. *Neuron*, 43, 623–631.
- Morris, C. A., Demsey, S. A., Leonard, C. O., Dilts, C., & Blackburn, B. L. (1988). The natural history of Williams syndrome: Physical characteristics. *Journal of Paediatrics*, 113, 318L 326.
- Napier, J. R. (1956). The prehensile movements of the human hand. *The Journal of Bone and Joint Surgery*, 38B, 902–913.
- Porter, M. A., & Coltheart, M. (2005). Cognitive heterogeneity in Williams syndrome. *Developmental Neuropsychology*, 27, 275L 306.
- Porter, M. A., Coltheart, M., & Langdon, R. (2008). Theory of Mind in Williams syndrome assessed using a non-verbal task. *Journal of Autism and Developmental Disorders*, 38, 806–814.
- Riby, D. M., & Hancock, P. J. B. (2008). Viewing it differently: Social scene perception in Williams syndrome and autism. *Neuropsychologia*, 46, 2855–2860.
- Rizzolatti, G., Fadiga, L., Gallese, V., & Fogassi, F. (1996). Premotor cortex and the recognition of motor actions. *Cognitive Brain Research*, 3, 131L 141.
- Rizzolatti, G., & Sinigaglia, C. (2010). The functional role of the parieto-frontal mirror circuit: Interpretations and misinterpretations. *Nature Reviews Neuroscience*, 11, 1–12.
- Rochat, P. (2007). Intentional action arises from early reciprocal exchanges. *Acta Psychologica*, 124, 8–25.
- Rodgers, J., Riby, D. M., Janes, E., Connolly, B., & McConachie, H. (2011). Anxiety and repetitive behaviour in autism spectrum disorders and Williams syndrome: A cross-syndrome comparison. *Journal of Autism and Developmental Disorders*, doi:10.1007/s10803-011-1225-x
- Roid, G. H., & Miller, L. J. (2002). *Leiter-R, Leiter International Performance Scale-Revised*. Firenze: Giunti O.S.
- Santos, A., & Deruelle, C. (2009). Verbal peaks and visual valleys in theory of mind ability in Williams syndrome. *Journal of Autism and Developmental Disorders*, 39, 651–659.
- Saxe, R., Carey, S., & Kanwisher, N. (2004). Understanding other minds: Linking developmental psychology and functional neuroimaging. *Annual Review of Psychology*, 55, 87–124.
- Searcy, Y. M., Lincoln, A. J., Rose, F. E., Klima, E. S., Bavar, N., & Korenberg, J. R. (2004). The relationship between age and IQ in adults with Williams syndrome. *American Journal on Mental Retardation*, 109, 231–236.
- Semell, S., & Rosner, S. (2003). *Understanding Williams syndrome: Behavioural patterns and interventions*. Mahwah, NJ: Lawrence Erlbaum Associates, Inc.
- Sommerville, J. A., Hildebrand, E. A., & Crane, C. C. (2008). Experience matters: The impact of doing versus watching on infants' subsequent perception of tool use events. *Developmental Psychology*, 44, 1249–1256.
- Sommerville, J. A., & Woodward, A. L. (2005). Pulling out the intentional structure of human action: The relation between action production and processing in infancy. *Cognition*, 95, 1–30.
- Sommerville, J. A., Woodward, A. L., & Needham, A. (2005). Action experience alters 3-month-old infants' perception of others' actions. *Cognition*, 96, B1–B11.
- Spunt, R. P., Satpute, A. B., & Lieberman, M. D. (2010). Identifying the what, why and how of an observed action: An fMRI study of mentalizing and mechanizing during action observation. *Journal of Cognitive Neuroscience*, 23, 63–74.
- Stefanini, S., Bello, A., Volterra, V., & Carlier, M. (2008). Types of prehension in children with Williams-Beuren syndrome: A pilot study. *European Journal of Developmental Psychology*, 5(3), 358–368.
- Stella, G., Pizzoli, C., & Tressoldi, P. E. (2000). *Peabody. Test di Vocabolario Ricettivo*. Torino: Omega Edizioni.
- Stromme, P., Bjornstad, P. G., & Ramstad, K. (2002). Prevalence estimation of Williams syndrome. *Journal of Child Neurology*, 17, 269–271.
- Sullivan, K., & Tager-Flusberg, H. (1999). Second-order belief attribution in Williams syndrome: Intact or impaired. *American Journal of Mental Retardation*, 104, 523–532.
- Tager-Flusberg, H., Plesa Skwerer, D., & Joseph, R. M. (2006). Model syndromes for investigating social cognitive and affective neuroscience: A comparison of autism and Williams syndrome. *Social Cognitive & Affective Neuroscience*, 1, 175–182.
- Tager-Flusberg, H., & Sullivan, K. (2000). A componential view of theory of mind: Evidence from Williams syndrome. *Cognition*, 76, 59–89.
- Tager-Flusberg, H., Sullivan, K., & Boshart, J. (1997). Executive functions and performance on false belief tasks. *Developmental Neuropsychology*, 13, 487L 493.
- Udwin, O., Howlin, P., Davies, M., & Mannion, E. (1998). Community care for adults with Williams syndrome: How families cope and the availability of support networks. *Journal of Intellectual Disability Research*, 42, 238–245.
- Umiltà, M. A., Escola, L., Intskirveli, I., Grammont, F., Rochat, M., Caruana, F., et al. (2008). How pliers become fingers in the monkey motor system. *Proceedings of the National Academic of Sciences of the United States of America*, 105, 2209–2213.
- Vicari, S., Bates, E., Caselli, M. C., Pasqualetti, P., Gagliardi, C., Tonucci, F., et al. (2004). Neuropsychological profile of Italians with Williams syndrome: An example of a dissociation between language and cognition? *Journal of International Neuropsychology Society*, 10, 862–876.
- Vicari, S., Carlesimo, G., Brizzolara, G., & Pezzini, G. (1996). Short-term memory in children with Williams syndrome: A reduced contribution of lexical-semantic knowledge to word span. *Neuropsychologia*, 34, 919L 925.
- Vicari, S., Caselli, M. C., Gagliardi, C., Tonucci, F., & Volterra, V. (2002). Language acquisition in special populations: A comparison between Down and Williams syndromes. *Neuropsychologia*, 40, 2461–2470.
- Volterra, V., Capirci, O., Pezzini, G., Sabbadini, L., & Vicari, S. (1996). Linguistic abilities in Italian children with Williams syndrome. *Cortex*, 32, 663L 677.
- Woodward, A. L., & Sommerville, J. A. (2000). Twelve-month-old infants interpret action in context. *Psychological Science*, 11, 73–77.
- Woodward, A. L., Sommerville, J. A., Gerson, S., Henderson, A. M. E., & Buresh, J. S. (2009). The emergence of intention attribution in infancy. In B. Ross (Ed.), *The psychology of learning and motivation*. Academic Press.
- Zheng, J. Z., De La Rosa, S., & Dollar, A. M. (2011). An investigation of grasp type and frequency in daily household and machine shop tasks. In *Proceedings of the 2011 IEEE International Conference on Robotics and Automation (ICRA)* Shanghai, China, May 9–13, (pp. 4169–4175).