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Abstract

This paper provides the first assessment of theory of mind, i.e. the ability to reason about mental states, in adult patients with congenital heart disease (CHD). Patients with CHD and matched healthy controls were administered classical theory of mind tasks and a semi-structured interview which provides a multi-dimensional evaluation of theory of mind (Th.o.m.a.s.). The patients with CHD performed worse than the controls on the Th.o.m.a.s., whereas they did as well as the control group on the classical theory of mind tasks. These findings provide the first evidence that adults with CHD may display specific impairments in theory of mind.

Keywords: adults; psychological distress; social cognitions; emotions; health psychology.

INTRODUCTION

Congenital heart disease (CHD) refers to a heterogeneous category of defects in the structure of the heart which are present from birth. ~~The improvement in diagnostic tools and surgery procedures from the early 1980s has led to a drastic reduction of the mortality rate in children born with this illness. The consequent significant increase of the so called "grown-up congenital heart disease" (GUCH) population has encouraged a transition in the guidelines for the management of the physical and psychological issues associated with CHD from paediatric to adult care (Baumgartner, Bonhoeffer, De Groot, de Haan, Deanfield et al., 2010).~~

From a psychological perspective, a large literature suggests that children with CHD are at higher risk of cognitive, behavioural and social difficulties. In particular, there seems to be a causal association between severity of the cardiac disorder, determined on the basis of medical variables such as cyanosis (i.e. excess of deoxygenated blood), palliative (versus corrective) surgery, or age at surgery, and decreased cognitive functioning, as reflected in lower IQ scores or academic achievement (for a recent meta-analysis see Karsdorp, Everaerd, Kindt & Mulder, 2007). However, the behavioural and social difficulties displayed by these patients do not appear to be related to factors which strictly depend on the specific physiological characteristics or on the severity of the heart disease (Kovacs, Sears & Saidi, 2005). By contrast, they seem related to environmental and emotive factors, such as the unpleasant experience of medical operations and hospital admissions during infancy, the feeling of isolation and social awkwardness in peer relationships for the forced restrictions in physical and social activities, the limitation in the development and expression of interpersonal and emotional skills determined by overprotective parental attitudes, and the body image issues caused by the heart surgery scars

(Cohen, Mansoor, Langut & Lorber, 2007; Cornett & Simms, 2013; Daliento, Mapelli & Volpe, 2006; Green, 2004; Kovacs et al., 2005).

A recent proposal aimed at providing a functional synthesis of the diverse behavioural and psychosocial difficulties observed in patients with CHD has been put forward by Bellinger (2008), who suggested that these individuals may be at higher risk of developing deficits in social cognition. In particular, the hypothesis is that children with congenitally malformed hearts, due to a number of neurological and/or psychological factors, may suffer from a deficit of "theory of mind" (ToM), i.e. the ability to reason about one's own and other people's mental states – such as desires, intentions and beliefs – and to use these mental states to explain and predict behaviour (Premack & Woodruff, 1978; Wimmer & Perner, 1983), also defined in literature as "mindreading" (Baron-Cohen, Leslie & Frith, 1985). Bellinger (2008) argued that a ToM deficit might, in principle, be the unifying underlying cause of the pragmatic language deficit, the symbolic skills impairment and the difficulty in identifying and describing internal states that have been observed in these patients, and that eventually determine their poor interpersonal adjustment.

~~Hence, the author invited researchers to direct their attention on the assessment of social cognitive abilities in patients with CHD, with the final aim to provide them with the support they need to achieve a satisfactory psychosocial functioning.~~

To our knowledge, only one study, carried out by Calderon and colleagues (2010), has empirically ~~to date responded to this call,~~ investigated~~ing~~ ToM in school-aged children with CHD and in healthy matched control participants. Children were presented with first-order (Wimmer & Perner, 1983) and second-order false belief tasks (Perner & Wimmer, 1985), to assess their understanding of, respectively,

another person's (false) belief (e.g. John has a false belief on an object location) and another's person belief embedded in yet another person's (false) belief (e.g. John has a false belief about Mary's belief on an object location) social interactions, and with a battery of tests to measure executive functioning (in particular, inhibition, working memory and planning). Children with CHD performed worse than control participants in both ToM tasks and in all the executive functioning tests, but only the ToM tasks and the inhibition test were reliable contributors to the difference between the children with CHD and the control group (Calderon, Bonnet, Courtin, Concordet, Plumet et al., 2010). This is an important study, because it identifies for the first time an impairment in ToM in patients with congenitally malformed hearts. However, it also presents an important limitation, as the authors themselves acknowledge, because it has been widely demonstrated that, in development, performance in inhibition tests is a strong predictor of accuracy in false belief tasks, as children have to inhibit their own knowledge of reality in order to report the (incorrect) believed state of affairs of another person (Carlston, Moses & Breton, 2002; Leslie, Friedman & German, 2004). Thus, the study by Calderon and colleagues (2010) cannot rule out the possibility that the children with CHD's failure in the theory of mind tasks might have originated from a difficulty in the inhibition of their knowledge (i.e. of their true belief), rather than from a genuine impairment in belief reasoning. Failure in similar tasks by a sample of adult participants would constitute a much more stringent test, since the current evidence suggests that in adulthood executive functioning does not play a necessary role in ToM reasoning (Apperly, Samson & Humphreys, 2009).

Furthermore, to provide a complete and detailed profile of this multifaceted function, employing also tasks other the classical ToM tasks may prove useful, because what exactly these tests measure and how their results generalize to the whole

ToM ability have been repeatedly questioned in the literature. For instance, some authors have highlighted that to reason on a false belief is more difficult than to reason on a true one (Barres & Johnson-Laird, 1997) and that the false belief task requires cognitive abilities other than mindreading, concluding that the false belief task it is not the most appropriate test of ToM (Bloom & German, 2000). Also, the classical ToM tasks focus on a specific and narrow facet of the ToM ability, namely third-person ToM (i.e. the ability to reason on another person's mental states), overlooking first-person ToM (i.e. the ability to reason on one's own mental states). Indeed, some studies have argued that ToM has a complex nature that cannot be reduced to an on-off or an all-or-nothing functioning and have hinted to the possibility of decomposing it into different aspects or components (Tirassa, Bosco & Colle, 2006a). In particular, Nichols and Stich (2002) argued that understanding the first- and the third-persons are different activities that are mediated by different processes and recruit knowledge of different types. Vogeley, Bussfeld, Newen, Herrmann, Happé et al. (2001) conducted an fMRI study on healthy subjects which empirically supported such distinction. The authors found that different brain circuits were recruited when participants assumed the first- and the third-person perspective. Another distinction which has been mostly overlooked by classical ToM tasks and is orthogonal to that between first- and third-person ToM, is that between egocentrism and allocentrism (Frith & de Vignemont, 2005). In the egocentric perspective, others' mental states are represented in relation to the self, while in the allocentric perspective others' mental states are represented independently from the self. Finally, literature in the developmental domain shows that not all mental states are expressed and comprehended with the same ease. For instance, children express earlier, and find easier to comprehend, desire with respect to belief (Wellman & Wooley, 1990).

All this considered, the present study wished to provide a wide investigation of theory of mind in the grown-up CHD population. For this reason the participants were administered, in addition to a battery of classical theory of mind tests, a recently developed semi-structured interview, Theory of Mind Assessment Scale (Th.o.m.a.s; Bosco, Colle, De Fazio, Bono, Ruberti & Tirassa, 2009, see also Castellino, Bosco, Marhall, Marshall & Veglia, 2011, Bosco, Capozzi, Colle, Marostica & Tirassa, 2013; Laghi, Cotugno, Cecere, Sirolli, Palazzoni, & Bosco, 2013), which provides a complete and detailed profile of ToM reasoning, without recurring to the false belief paradigm and directly investigating all the different aspects of a person's ability to mindread, i.e. first vs. second order ToM, first vs. third person ToM, egocentric vs. allocentric perspective. In detail we hypothesised that patients with CHD would show an impaired ToM ability when compared with healthy controls. However, we also expected that - focusing on CHD performance - some aspects of their ToM might be better preserved than others. In particular we hypothesised that: i) the performance of patients with CHD in first-person ToM might be significantly better than that in third-person ToM; and ii) the performance of patients with CHD in first order ToM tasks might be better than that in second order ToM tasks. Finally, for explorative purposes, we wished investigate iv) possible differences in patients' performance in ToM tasks requiring an allocentric vs. an egocentric perspective and in v) their ability to reason on different types of mental states, i.e. emotion, desire and belief.

METHODS

Participants

Patients with congenital heart disease were recruited from the Cardiology Divisions of two Italian hospitals. Inclusion criteria included age between 18 and 60 years, presence of complex CHD, surgical correction of the heart defect, to be a native speaker of Italian, willingness to participate to the research as expressed by reading the informative sheet and signing the informed consent form, and a normative range (equivalent scores of 3-4, indicating a performance at or above the 50th centile of normal range; Spinnler & Tognoni, 1987) in IQ as measured by Raven's *Standard Progressive Matrices* (SPM; Raven, 1992), working memory as measured by the *Wechsler Digit Span* test (Wechsler, 1981), and executive function as measured by the *Tower of London* test (Shallice, 1982). Exclusion criteria included major chromosome abnormalities and history of severe physical, neurological or psychiatric disorders seriously affecting cognitive and/or psychological functioning.

After reviewing the medical records of the databases of the two Cardiology Divisions, all the patients who met the selection criteria were contacted by a psychologist and were explained the aim of the study and the procedure. Twenty-five patients accepted to participate and, of these, nine patients had to be subsequently excluded because their IQ, working memory and/or executive function scores fell outside the normative range, thus leaving sixteen patients (6 F, 10 M; mean age 29.1 years, age range 18-57 years; mean education 11.2 years, education range 8-17 years). In addition, sixteen healthy control volunteers matched to the study group for sex, age, and years of formal education (6 F, 10 M; mean age 29.2 years, age range 18-54 years; mean education 11.3 years, education range 8-17 years) were recruited from the general population. Written informed consent was obtained from all the participants. Table 1 summarizes the demographic and clinical characteristics of the study participants. The study was performed in accordance with the ethical standards

of the 1964 Declaration of Helsinki and was approved by the Ethical Committee of the Hospital.

Procedures

Participants were tested individually on the following measures. The order in which the measures were administered was counterbalanced across participants within each group.

[Table 1 about here]

Measures

Classical theory of mind (ToM) tasks

The following measures (hereafter referred to as ToM tasks) were used to assess the theory of mind ability of both the patient and the control group.

Strange Stories task. The Strange Stories task (Happé, 1994) is an advanced or 'high level' theory of mind task. Six Strange Stories (i.e. banana, boat, gloves, miss Bianchi, soldier, tennis bat) were selected from the adapted set of stories by Happé and colleagues (Happé, Brownell, & Winner, 1999). The scoring procedure follows that described by Happé (1994), with a score of 0 being assigned to each incorrect answer and a score of 1 to each correct one.

Second-order ToM task. The burglar story (Happé & Frith, 1994) and the ice-cream van story (Baron-Cohen, 1989) were administered to the participants. Both these tests adopt a similar strategy, called the 'double-bluff story', where the subject must attribute to a story character's a false belief about another character's belief; the correct answer requires the ability to understand second-order belief. The scoring

procedure follows that described by the authors, with a score of 0 being assigned to each incorrect answer and a score of 1 to each correct one.

Theory of Mind Assessment Scale (Th.o.ma.s.)

In addition to the classical ToM tasks, it was also administered the Theory of Mind Assessment Scale (Th.o.m.a.s.; Bosco et al., 2009), a semi-structured interview developed to assess several components of theory of mind. This measure provides a complete and detailed profile of this cognitive function and is described in more detail in Bosco et al. (2009). Th.o.m.a.s. consisting s of 39 open-ended questions that leave the interviewees free to express and articulate their thoughts. The interviewer is trained to repeat and explain the questions to the participant on request at any time during the assessment, and this procedure allows to control for task-related demands, such as memory, language or attention.

The questions are organized into four scales, each focusing on one of the knowledge domains in which a person's theory of mind may manifest itself.

Scale A, I–Me. It investigates the interviewee's knowledge of her own mental states. The viewpoint of the questions is centred on the interviewee (I) reflecting on her own mental states (Me) (e.g., "Do you ever experience emotions that make you feel good?"). This scale investigates first-person ToM in an egocentric perspective.

Scale B, Other–Self. It investigates the knowledge that, according to the interviewee, other persons have of their own mental states, independently of the subject's perspective. The viewpoint of the questions is centred on the other persons (Other) reflecting on their own mental states (Self) (e.g., "Do other people try to fulfil their wishes?"). This scale investigates third-person ToM in an allocentric perspective.

Scale C, I–Other. It investigates the interviewee's knowledge of the mental states of other persons. The viewpoint of the questions is centred on the interviewee (I) reflecting on the others' mental states (Other) (e.g., "Do you notice when other people feel good?"). This scale is similar to scale B in that they both investigate third-person ToM; however, while the perspective there is centred on the other person, here it is centred on the interviewee. In other words, here the subject is asked to take an egocentric perspective.

Scale D, Other–Me. It investigates the knowledge that, from the interviewee's point of view, other people have of her mental states. The viewpoint of the questions is centred on the other persons (Other) reflecting on the mental states of the interviewee (Me) (e.g., "Do other people notice when you feel good?"). This scale can be compared with a second-order ToM task, in that the abstract form of the questions is: "What do you think that the others think that you think?".

Each scale is further divided into three subscales, which explore, respectively, the dimensions of *a~~A~~wareness,* i.e. the interviewee's ability to perceive and differentiate beliefs, desires and emotions in herself and in other people ~~Relation and Realization of mental states:~~

~~*Awareness*. It investigates the interviewee's ability to perceive and differentiate beliefs, desires and emotions in herself and in other people~~. ~~Recognizing different types of mental states is a necessary precondition for understanding their links and causal relations with one another and with the external world.~~

r~~R~~elation, ~~.~~ ~~It investigates~~i.e. the interviewee's ability to recognize causal relations between different mental states and between them and the resulting behaviours., and ~~For example: "When you feel bad, do you feel you understand why?". Being capable to connect and to integrate different mental states and to~~

understand their reciprocal relations and bi-directional connections with perceptions and actions is necessary to draw up an explanatory theory of the mind and of the social world.

rRealization. It investigatesi.e.., the interviewee's ability to adopt effective strategies to achieve a desired state. For example: "Do you succeed in getting what you want? How?". To act adaptively requires not only to have a theory of the causal relations between mental states and between the mental states and the world, but also to know how to use this knowledge to appropriately and successfully affect the mental states and the behaviour of one's own and of the other people.

Based on current theorizing on the most important types of mental states that an agent's cognitive architecture has to comprise (Tirassa & Bosco, 2008; Tirassa, Bosco & Colle, 2006a; 2006b; Bosco, Colle & Tirassa, 2009), the questions of the Th.o.m.a.s. interview focus on the interviewee's perspective on epistemic states (such as knowledge and beliefs), volitional states (such as desires and intentions) and positive and negative emotions.

All the Th.o.m.a.s. interviews were tape-recorded and then transcribed. The transcripts were rated by two independent judges, who had not participated in the interviewing phase and were blind as to whether the participants were in the CHD or in the control group. Each judge assigned a score from 0 to 4 to every answer, according to the rating criteria, and then inserted the score in the relevant cell of the correction grid (the whole structure of the interview, the coding criteria and the scoring grid may be found in Bosco et al., 2009). The judges reached a satisfactory and significant level of inter-rater reliability in terms of their initial judgments (Cohen's K test: $K = .678$, $p < .001$). For the final score assignment, the judges discussed each item upon which they disagreed until full agreement was reached.

Data Analysis

ANOVAs were conducted to investigate the patterns of scores of the patients with CHD and of the controls on both the classical ToM tasks and the Th.o.m.a.s. In addition, when appropriate, Bonferroni corrected t-tests were performed to investigate patients and controls' performance on each ToM task and on each of the Th.o.m.a.s. scales, subscales and dimensions.

RESULTS

Classical ToM tasks: comparison between patients with CHD and controls

Figure 1 shows the mean total scores for the patients and the controls on each of the classical ToM tasks (the six Strange Stories and the two second order stories).

A mixed-model ANOVA with a two-level between-subjects factor (group: patients vs. controls) and a two-level within-subjects factor (classical ToM task: Strange Stories and second order stories) was applied to investigate whether the patients' performance in the two classical ToM tasks was worse than that of the controls. The results revealed only a main effect of the ToM task ($F_{(1,30)} = 19.81$, $p < .001$, $\eta^2 = .40$) (all other $F < .04$, all $p > .835$), indicating that both the patients and the controls performed worse on the second order ToM task than on the Strange Stories task, but that there was no difference in performance between the two groups.

Th.o.m.a.s.: comparison between patients with CHD and controls

Overall, patients performed worse than controls on the Th.o.m.a.s. ($t_{(30)} = 2.95$, $p = .009$). Figure 2 shows the mean scores for the patients and the controls on

each individual Th.o.m.a.s. scale (A, B, C and D). A mixed-model ANOVA was performed with a two-level between-subjects factor (group: patients vs. controls) and a four-level within-subjects factor (Th.o.m.a.s. scale: A, I-Me; B, Other-Self; C, Me-Other; D, Other-Me). There was a main effect of group ($F_{(1,30)} = 8.70$, $p = .006$, $\eta^2 = .23$); overall, patients performed worse than control participants on the Th.o.m.a.s. scales. Furthermore, there was a main effect of scale ($F_{(3,90)} = 4.26$; $p = .007$; $\eta^2 = .12$), and the group*scale interaction was also significant ($F_{(3,90)} = 6.67$; $p < .001$, $\eta^2 = .18$). To explore this result, a series of post-hoc Bonferroni corrected (alpha ≤ .012) t-tests revealed that patients' performance was significantly worse than controls on scale B ($t_{(30)} = 3.68$, $p = .002$) and on scale C ($t_{(30)} = 3.43$, $p = .004$), which both assess third-person ToM (the first from an allocentric perspective, the second from an egocentric perspective), while there was no difference between the two groups in scale A, investigating first-person ToM ($t_{(30)} = .52$, $p = .610$), and in scale D, which investigates second order ToM ($t_{(30)} = 1.78$, $p = .092$).

~~Figure 3 shows the mean total score for both groups on the three Th.o.m.a.s. subscales (Awareness, Relation and Realization). A mixed-model ANOVA was performed with a two-level between-subjects factor (group: patients vs. controls) and a three-level within-subjects factor (subscale: Awareness, Relation, Realization). The analysis revealed a main effect of group ($F_{(1,30)} = 9.62$, $p = .004$, $\eta^2 = .24$), indicating that the patients obtained lower overall scores than the controls. There was also a significant main effect of scale ($F_{(2,60)} = 6.40$, $p = .003$, $\eta^2 = .18$), indicating that the participants' scores varied according to the subscale involved. The group*subscale interaction was marginally significant ($F_{(2,60)} = 2.93$, $p = .061$, $\eta^2 = .09$). To explore this result, a series of post-hoc Bonferroni corrected (alpha ≤ .017) t-tests revealed that the patients' performance was significantly worse than that of the controls on the~~

~~subscales Awareness ($t_{(30)}$ = 3.37, p = .004) and Realization ($t_{(30)}$ = 2.83, p = .010) and, marginally, also on the scale Relation ($t_{(30)}$ = 2.24, p = .039, with alpha ≤ .017).~~

Figure 34[FB1] shows the mean total score for the patient and control participants for each kind of mental state (belief, desire, positive and negative emotion). A mixed-model ANOVA was performed with a two-level between-subjects factor (group: patients vs. controls) and a four-level within-subjects factor (mental state: belief, desire, positive emotion, negative emotion). This analysis showed a main effect of group ($F_{(1,30)}$ = 9.06, p = .005, η^2 = .23), suggesting that overall the patients' performance was worse than that of the controls. There was also a main effect of mental state ($F_{(3,90)}$ = 8.89, p < .001, η^2 = .23) and the group*mental state interaction was also significant ($F_{(3,90)}$ = 5.33, p = .002, η^2 = .15). To explore this result, a series of post-hoc Bonferroni corrected (alpha ≤ .012) t-tests revealed that the patients' performance was significantly worse than that of the controls on the mental states belief ($t_{(30)}$ = 3.25, p = .005), positive emotion ($t_{(30)}$ = 2.88, p = .010) and, marginally, also negative emotion ($t_{(30)}$ = 2.15, p = .045, with alpha ≤ .012), while there were no differences for desire ($t_{(30)}$ = 1.17, p = .255).

Th.o.m.a.s.: performance within patients with CHD

To better characterize the performance of the patients with CHD in the Th.o.m.a.s. scale, a repeated-measures ANOVA with a four-level within-subjects factor (Th.o.m.a.s. scale: A, I-Me; B, Other-Self; C, Me-Other; D, Other-Me) was performed on the patient group (see Figure 2). The effect of scale was significant ($F_{(3,45)}$ = 5.59, p = .002), and Bonferroni corrected (alpha ≤ .050) multiple comparisons showed that patients performed better on scale A, which assesses first-person ToM, than on scale B (p = .016), which assesses third-person ToM, and on scale D ($t_{(15)}$ =

3.58, $p = .017$), which assesses second-order ToM (all other $p > .154$). The same analysis on the control group also showed a significant effect of scale ($F_{(3,45)} = 4.67$, $p = .006$), but no differences emerged in the multiple comparisons among the Th.o.m.a.s. scales (all $p > .065$).

~~A repeated-measures ANOVA with a three-level within-subjects factor (Th.o.m.a.s. subscale: Awareness, Relation, Realization) was then performed on the patient group (see Figure 3). The effect of subscale was significant ($F_{(2,30)} = 4.60$, $p = .018$), but no differences emerged in the multiple comparisons among the Th.o.m.a.s. subscales (all $p > .076$). The same analysis on the control group also showed a significant effect of scale ($F_{(2,30)} = 5.24$, $p = .011$), and Bonferroni corrected (alpha ≤ .050) multiple comparisons showed that healthy participants performed marginally better in the Awareness than in the Relation subscale ($p = .056$) (all other $p > .111$).~~

Finally, a repeated-measures ANOVA with a four-level within-subjects factor (Th.o.m.a.s. mental state: belief, desire, positive and negative emotion) was performed on the patient group (see Figure 3). The effect of mental state was significant ($F_{(3,45)} = 7.76$, $p < .001$), and Bonferroni corrected (alpha ≤ .050) multiple comparisons showed that patients performed better relative to the mental state desire than to the mental states belief ($p = .024$) and positive emotion ($p = .011$) (all other $p > .097$). The same analysis on the control group did not reveal any significant difference ($F_{(3,45)} = 2.51$, $p = .071$).

DISCUSSION

The aim of the present study was to assess ToM in a group of adult patients with CHD using a battery of classical ToM tasks (Baron-Cohen, 1989; Happé, 1994;

Happé & Frith, 1994), in addition to a more recent clinical tool, the Th.o.m.a.s. (Bosco et al., 2008), which is a semi-structured interview allowing to investigate several facets of the ToM ability, i.e. first-person vs. third-person, first order vs. second order, egocentric vs. allocentric.

In line with the recent theoretical proposal of Bellinger (2008), suggesting that patients with CHD are at higher risk of developing deficits in ToM, results showed that patients with CHD performed worse than controls overall in the Th.o.m.a.s., and in particular in the two scales investigating third-person ToM, i.e. the ability to understand another's person mental states, both from an allocentric (scale B) and from an egocentric (scale C) perspective. By contrast, no differences were found between patients and controls in their performance in scale A, investigating first-person ToM. These results are in line with previous work showing that schizophrenic patients (Bosco et al., 2008) and sex offenders (Castellino et al., 2011) performed worse on the third-person than on the first-person Th.o.m.a.s. scales. In addition, in line with these results, it was found that the patients with CHD, but not the controls, performed better in scale A rather than in scale B. Taken together, these results seem to support the hypothesis of Goldman (1993) that human beings can better reason about their own mental states than about those of the others.

By contrast no significant difference emerged in patients' performance in the comparison of scale B, investigating third person ToM from and allocentric perspective, with scale C, investigating third person ToM from an egocentric perspective, testifying that such different perspective is not a crucial mindreading dimension for patients with CHD.

The results also showed an almost significant difference between patients and controls' performance in scale D, investigating second order ToM. The lack of a fully

significant effect may be due the small number of participants in our clinical sample. Indeed the patients, but not the controls, performed worse on scale D, investigating second order ToM, than on scale A, investigating first order ToM. Such result is in line with the findings of the study by Calderon et al. (2010), showing that patients with CHD were more impaired in second level ToM than in first level ToM.

~~About the three subscales investigated by the Th.o.m.a.s. interview (Awareness, Relation, Realization), it was found that the patients with CHD performed worse than controls in all of them. In other words, the patients had difficulties in differentiating beliefs, desires and emotions in themselves and in other people, in understanding the causal relations between different mental states and between mental states and behaviour, and in adopting effective strategies to achieve a desired state. Mindreading difficulties may constitute one of the factors underlying the poor psychosocial adjustment observed in patients with CHD, who in a number of studies have been shown to display more psychological and emotional distress (Brandhagen, Feldt & Williams, 1991; Horner, Liberthson & Jellinek, 2000; Van Rijen, Utens, Roos-Hesselink, Meijboom, van Domburg, et al., 2003) and more depression and anxiety symptoms (Bromberg, Beasley, D'Angelo, Landzberg & DeMaso, 2003; Popelova, Slavik & Skovranek, 2001) compared to healthy controls.~~

Finally, about the specific mental states investigated by the Th.o.m.a.s. (belief, desire, positive and negative emotion), our results revealed that the patients' performance was worse compared to the controls in the questions investigating beliefs and emotions, while there were no differences for desires. In particular, the patients – but not the controls – performed worse on the questions investigating belief and positive emotion than on the questions investigating the mental state of desire. This result is in line with the literature on the development of mindreading abilities, which

shows that children find beliefs harder to understand than desires, and that the ability to handle beliefs is developed later than the ability to reason about other epistemic mental states (Wellman & Wolley, 1990). Mindreading difficulties may constitute one of the factors underlying the poor psychosocial adjustment observed in patients with CHD, who in a number of studies have been shown to display more psychological and emotional distress (Brandhagen, Feldt & Williams, 1991; Horner, Liberthson & Jellinek, 2000; Van Rijen, Utens, Roos-Hesselink, Meijboom, van Domburg, et al., 2003) and more depression and anxiety symptoms (Bromberg, Beasley, D'Angelo, Landzberg & DeMaso, 2003; Popelova, Slavik & Skovranek, 2001) compared to healthy controls.

For what concerns the classical ToM tasks, i.e. the Strange Stories (Happé, 1994) and the second order ToM tasks (Baron-Cohen, 1989; Happé & Frith, 1994), no difference was found in the performance of patients with CHD and controls. A possible reason is that, as mentioned in the Introduction, the classical ToM tasks, which are based on the false belief paradigm, may not be the most appropriate test of ToM, because they do not specifically and exclusively tap ToM abilities (Barres & Johnson-Laird, 1997; Bloom & German, 2000). Also, having been initially developed for children, they provide a coarse measure of ToM and they may not be especially suited to be used with adults. By contrast, Th.o.m.a.s. has been developed specifically for adult persons, and thus appears a more suitable and sensitive clinical tool to highlight mindreading deficits in adult patients with CHD.

A last consideration relates to the fact that a third of our initial sample of patients with cyanotic CHD had to be excluded from the study because it fell outside the normative range for IQ, working memory and/or executive function. These data

suggest that CHD may be associated with serious cognitive difficulties, but also that, after controlling for general cognitive abilities, patients with CHD may still display mindreading impairments. Indeed, the present study demonstrates for the first time the presence of a deficit of ToM in adults with CHD. An interesting question for future studies is whether such ToM difficulties depend upon the specific physiological characteristics or the severity of the heart disease, or rather upon environmental and emotive factors.

These results are consistent with the need to develop interdisciplinary programs of assistance for patients with CHD, in consideration of the different aspects that contribute to determine their quality of life (Chiavarino, Rabellino, Cavallero, Palumbo, Bergerone, Gaita, et al., 2012; Daliento et al., 2006; Green, 2004; Kovacs et al., 2005). In particular, they suggest that children and adults with CHD may benefit from cognitive interventions aimed at improving their theory of mind ability. Theory of mind plays a crucial role in setting and maintaining social relationships; it therefore appears important to understand exactly which aspects of this cognitive ability are compromised in patients with CHD, and with which degree of severity, in order to allow an efficient rehabilitation process with positive fallouts on their quality of life.

ACKNOWLEDGMENTS

The research was supported by Ministero Italiano dell'Università e della Ricerca: PRIN project "New perspectives about Theory of the Mind: from early infancy to adulthood" project code 2008N9KF5K. We thank all the patients for their kind participation.

NO CONFLICTING INTERESTS

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TABLES

	Patients with CHD (N=16)	**Healthy controls (N=16)**
Sex (%)	37.5 F – 62.5 M	37.5 F – 62.5 M
Mean Age, in years (SD)	29.1 (11.9)	29.2 (12.4)
Mean Education, in years (SD)	11.2 (2.9)	11.3 (3.2)
Diagnosis (%)		
Right ventricular tract obstruction	31.2	

Pulmonary valve disease	50.0
Tetralogy of Fallot	18.8
Transposition of the great arteries	
Mean Age of first surgery, in months (SD)	16.6 (16.1)

Table 1. Demographic and clinical characteristics of the study participants.

FIGURES

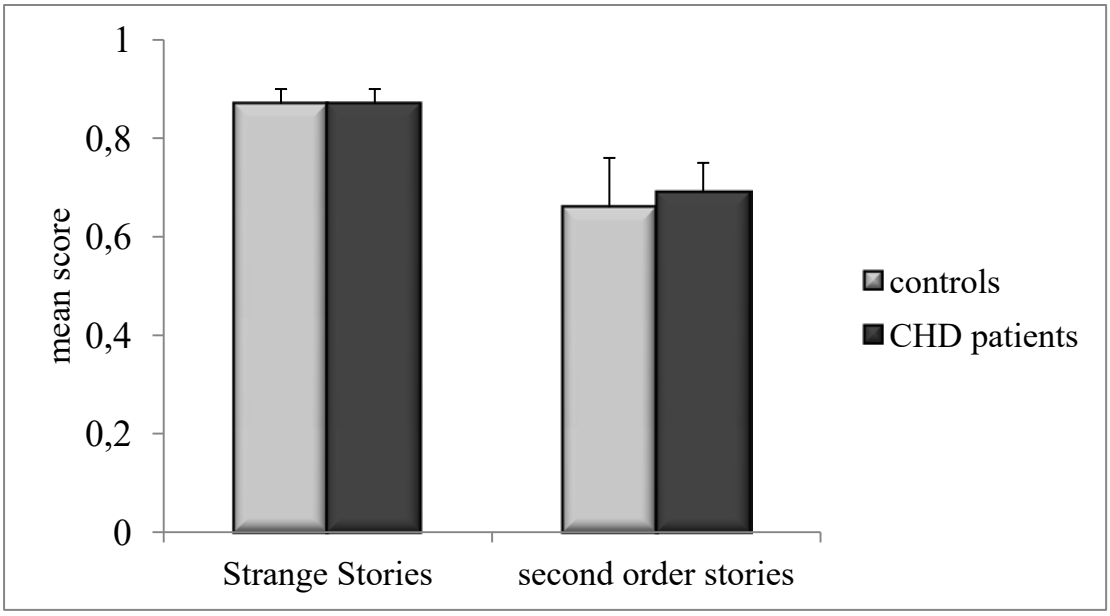


Figure 1. Patients with CHD vs. controls: mean total scores in the Strange Stories and second order stories (range: 0–1). Error bars represent standard errors of the mean.

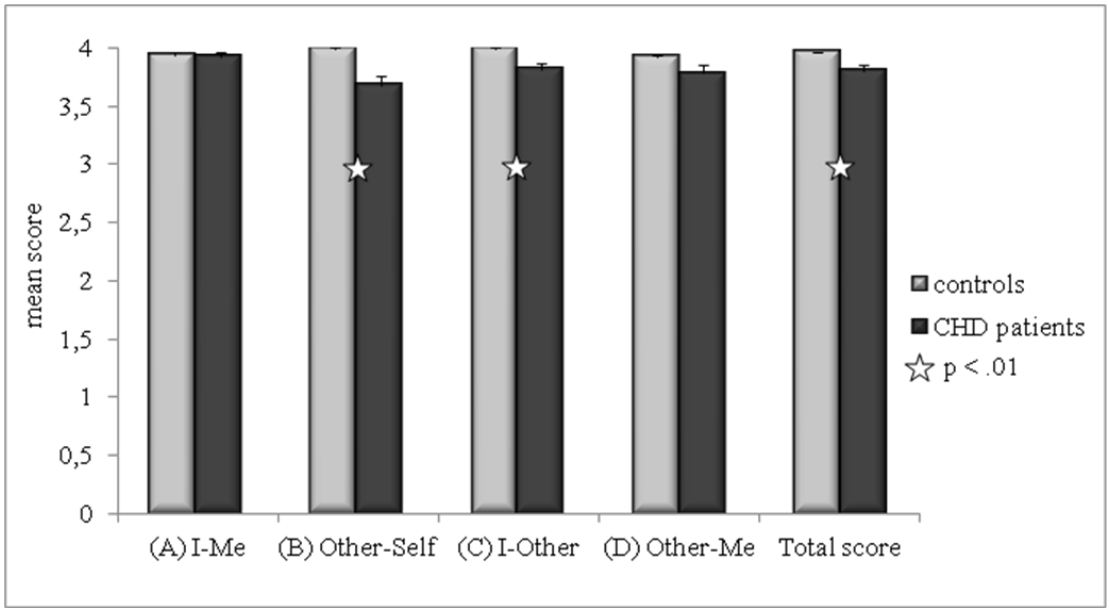
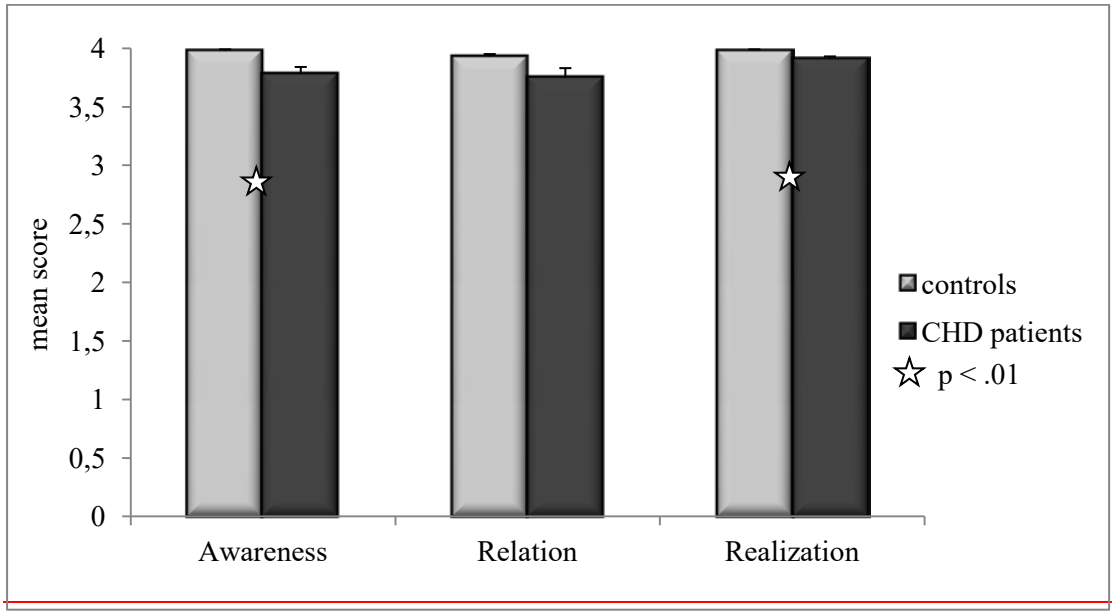


Figure 2. Patients with CHD vs. controls: mean total scores in the Th.o.m.a.s. and mean scores in the individual scales (range: 0–4). Error bars represent standard errors of the mean.



~~**Figure 3.** Patients with CHD vs. controls: mean total scores in the individual subscales (range: 0–4). Error bars represent standard errors of the mean.~~

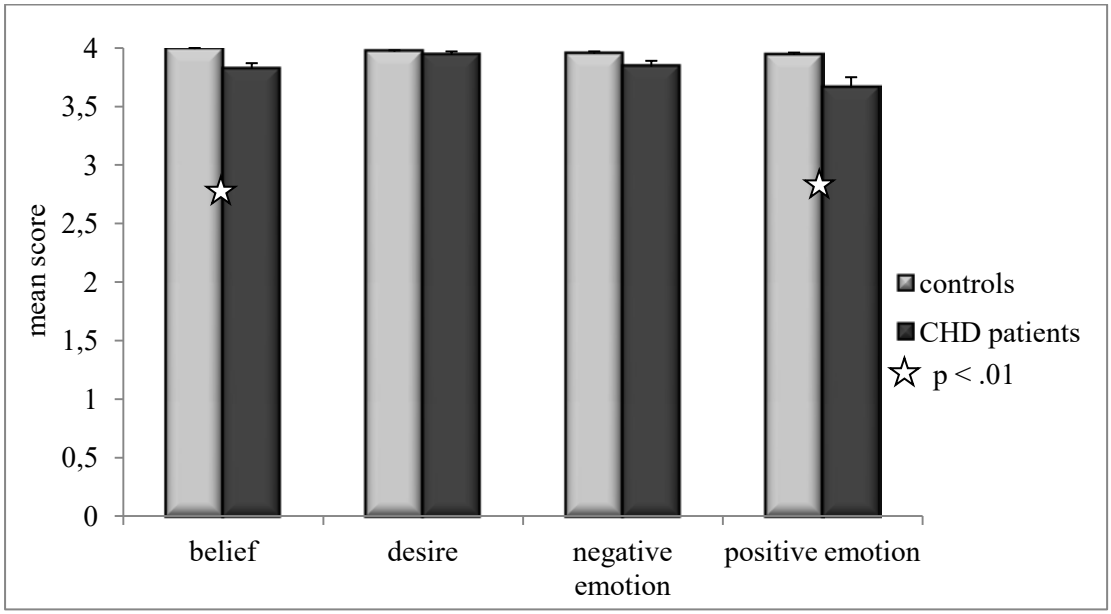


Figure 34. Patients with CHD vs. controls: mean total scores in the individual mental states (range: 0–4). Error bars represent standard errors of the mean.